The effects of a pragmatic community exercise programme in adolescents and young adults with cerebral palsy

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Abstract

Evidence suggests that the walking ability of people with cerebral palsy (CP) often deteriorates in early adulthood. This is especially problematic as the health care for young people with disabilities changes considerably over the course of the transition from paediatric to adult health care services. This suggests the importance of providing young people with the appropriate tools for managing their disability on a largely independent basis if they hope to reduce or delay this physical decline. One such strategy is the introduction of young people to an exercise programme which may be carried out independently of the health care system. Therefore, the main aim of this thesis was to investigate the feasibility and effectiveness of an 18-week pragmatic community exercise programme in adolescents and young adults with CP. Acknowledging the importance of assessing the effects of exercise studies using outcome measures (OMs) which are reliable, valid and responsive to change, a secondary aim of this thesis was to synthesise the psychometric evidence for the measures of gait quality and walking performance currently used for adolescents and young people with CP through a systematic review. Additional psychometric evidence for measures commonly used to assess the efficacy of exercise interventions was provided by a test-retest reliability study as part of this thesis.

A standardised quality checklist (COSMIN) was used in the systematic review (Study 1) to measure methodological quality. The strength of the evidence was rated using standardised guidelines. The synthesis of best evidence was scored according to the Cochrane criteria, which indicated that the reliability (inter-rater) of the Functional Mobility Scale was characterised by a 'strong' level of evidence. The evidence for the responsiveness for all OMs included in this review was rated as 'unknown'. Only one study reported on measurement error when reporting on reliability. In Study 2, test-retest reliability (Intraclass Correlation Coefficient (ICC) and Minimal Detectable Change) was calculated for physical function, habitual physical activity (HPA), quality of life (QoL) and self-esteem measures in a group of adolescents and young adults with CP (n=8) and their age-matched peers (n=14). The ICCs for physical function and HPA OMs ranged from moderate to good but were poor for the measures of QoL and self-esteem. The RCT (Study 3) showed no statistically significant improvement following the exercise programme in any of the OMs at 6 weeks (experimental n=9, control n=7). A small effect size (d=0.54) in favour of the experimental group was found for the Canadian Occupational Performance Measure (COPM). Considering the experimental group only, a statistically significant improvement was found for the COPM at 12 weeks (n=7, p=0.02) compared to the baseline. Feasibility issues were also identified. This study was limited by its small sample size.

This thesis contributes to the evidence base on pragmatic community exercise programmes for adolescents and young adults with CP and confirms the test-retest reliability (consistency) of physical function and HPA OMs commonly used to assess the efficacy of exercise interventions in CP.

Key words: adolescents, young adults, cerebral palsy, exercise, reliability, minimal detectable change, measurement error, psychometric properties physical activity, isometric strength, 3D gait analysis, Quality of Life, Self-esteem

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Abbreviations

Abbreviations used within the thesis

3DGA	Three-dimensional gait analysis
CONSORT	Consolidated Standards for Reporting Trials
COSMIN	COnsensus-based Standards for the selection of health Measurement Instruments
CP	Cerebral Palsy
EQUATOR	Enhancing the Quality and Transparency of health Records
HPA	Habitual physical activity
ICC	Intraclass coefficient
ICF	International Classification and Function, disability and health
Kg	Kilogramme
M	Metre
MCID	Minimal clinically important change
MDC	Minimal detectable change
Nm/kg	Newton metre /kilogramme (normalised torque)
OMs	Outcome measures
PIS	Participant information sheet
QoL	Quality of Life
Secs	Seconds
SEM	Standard error of measurement
WHO	World Health Organization

Abbreviations used for outcome measures within this thesis

10m SRT	10-metre shuttle run test
COPM	Canadian Occupational Performance Measure
FAQ	Gillette Functional Assessment Questionnaire
GMFM	Gross Motor Function Measure 66
GPS	Gait Profile Score
ISM	Isometric muscle strength
RSES	Rosenberg Self-Esteem Scale
SF-12	Short form 12 version 2
TUG	Timed up and go test

Publications/presentations arising from this thesis

Zanudin A, Van der Linden M, Jagadamma K, Mercer T, 2016. European Society Motion Analysis Congress (ESMAC). Habitual physical activity in adolescent and young adult with Cerebral Palsy and age-matched peers [Poster Presentation] Seville, 30thMarch 2016

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CHAPTER 1 INTRODUCTION

Cerebral palsy (CP) occurs as a result of developmental brain injuries during foetal development, birth or shortly after birth (Baxter et al. 2007). CP is a non-progressive neurological condition, but it often results in secondary musculoskeletal conditions including pain, fatigue, muscle weakness and decreased mobility, which can lead to diminished independence. Some of these secondary conditions are likely to worsen during early adulthood (Tosi et al. 2009). The incidence of CP in the UK is reported to be two in every 1000 live births, and there is an increased likelihood of CP in children with low birth weights (Surman et al. 2009). With advancement in medical technologies and effective management, more than 90% of the CP population survive to adolescence and adulthood (Cathels and Reddihough 1993, Cooley and American Academy of Paediatrics Committee on Children With Disabilities 2004b).

Adolescence is an important time in terms of the care of young people with a long-term health condition, and those with CP are no exception. Paediatric care for people with CP is generally quite comprehensive and involves multidisciplinary treatment; however, once patients reach a certain age, generally around 16–21 years (Binks et al. 2007, Wright et al. 2016), they are discharged from child health care. Since CP was traditionally viewed as a childhood ailment, health care for adults with CP is typically not as well established and services similar to those available to CP patients during childhood are lacking (Field et al. 2010, Horsman et al. 2010b, Ng et al. 2003, Zaffuto-Sforza 2005). This likely contributes to the reported decrease in utilisation and exposure to health care services after patients reach adolescence, leave school and enter adulthood (Ng et al. 2003, Hilberink et al. 2007, Stevenson et al. 1997). This type of situation is common despite adults with CP experiencing problems such as decreased mobility (Jahnsen et al. 2004a, Liptak 2008), loss of walking ability (Bottos et al. 2001, Murphy

et al. 1995) and a high prevalence of pain and join deformities (Hilberink et al. 2007). This suggests the importance of providing patients with the right tools to self-manage their condition if they hope to reduce or delay the physical decline which occurs with ageing and which also occurs earlier in patients with CP than in the population without CP (Jahnsen et al. 2004a, Horsman et al. 2010a). Thus, strategies to help young people with CP to self-manage this decline in function are warranted. One such strategy is to introduce young people to an exercise programme, which can be followed without the health care system that is often lacking in resources.

A number of studies have examined the implementation of exercise programmes for people with CP in an attempt to improve muscle strength and other outcomes of physical fitness. Many have hypothesised that an increase in the physical fitness (International Classification of Function (ICF) (World Health Organisation 2008) – body structures and function level) of people with CP could lead to an improvement in their gait quality, walking performance, mobility (the ability of an individual to move about in the environment), gross motor function, habitual physical activity, self-esteem or quality of life (Dodd et al. 2003, Engsberg et al. 2006, Fowler et al. 2010, McNee et al. 2009, Lee et al. 2008, Morton et al. 2005, Liao et al. 2007, Patikas et al. 2006, Unger et al. 2006, Salem and Godwin 2009, Unnithan et al. 2007, Verschuren et al. 2007, Andersson et al. 2003, Debuse et al. 2009, Ballaz et al. 2010, Eek et al. 2008, Slaman et al. 2015, Bania et al. 2016). Most review articles in this area seem to support the idea of increased muscle strength and, in some cases, gross motor function, as a result of exercise interventions (Darrah et al. 1998, Dodd et al. 2002, Jeglinsky et al. 2010, Martin et al. 2010, Mockford and Caulton 2008, Verschuren et al. 2008a, Park and Kim 2014). However, closer evaluation reveals a number of uncertainties. These systematic reviews report a limited number of studies, which are often of low-quality study design, particularly those studies focusing on adolescents and young adults (Jeglinsky et al. 2010). In addition, Scianni et al. (2009) performed a meta-analysis of strength training in children and adolescents with CP that included only randomised control trials

(RCTs). Based on the available data, they concluded that strengthening interventions had no effect on strength or walking speed and only a small statistically significant, but not clinically meaningful, effect on Gross Motor Function Measure (GMFM). Only five studies were available for inclusion in the meta-analysis, and the authors acknowledged that factors such as insufficient intensity or duration of strength training may have contributed to these results. Only two of those five studies included only adolescents; Unger et al. (2006), which included participants aged 13–19 years, and Unnithan et al. (2007), which included participants aged 14–18 years.

In order to evaluate the efficacy of an exercise intervention, the clinician or researcher needs to select outcome measures (OMs) which are valid, reliable and responsive to change as well as practical and appropriate for the research question(s) to be answered. As a result, there has been also growing interest in exploring the level of evidence of psychometric properties of OMs used in the population with CP, as evidenced by the number of systematic reviews published recently. Thus far, however, no clear consensus has been reached within clinical groups in terms of which OM(s) to use when working on exercise prescription for individuals with CP. Further, the evidence of psychometric properties of these OMs is conflicting and, in some cases, remains unknown.

To summarise, there is ambiguity in the literature regarding the feasibility and effectiveness of exercise programmes in adolescents and young adults with CP, which may be attributable to the low-quality design and inappropriate exercise programme characteristics (e.g. insufficient frequency of the exercise sessions and/or duration of exercise programme) applied in the previous studies. Secondly, considering the importance of using psychometrically sound OMs to evaluate the effectiveness of a given exercise programme, it is important to explore the psychometric properties of the OMs in this population. Therefore, the overall aims of this thesis are:

1) To examine the level of psychometric evidence for measures of gait quality and walking performance used for adolescents and young adults with CP (Study 1).

2) To investigate the reliability (i.e. consistency and agreement) of measures of physical function, habitual physical activity, quality of life and self-esteem used for adolescents and young adults with CP (Study 2).

3) To investigate the physical function, habitual physical activity, quality of life and self-esteem in young people with CP and healthy controls (Cohort study).

4) To investigate the feasibility and effectiveness of an 18-week pragmatic community exercise programme in adolescents and young adults with CP (Study 3).

1.1 Contribution to knowledge

In undertaking the studies in this thesis, additional knowledge will be added to the growing body of literature surrounding exercise interventions for young people with CP. The impact of a pragmatic community exercise programme for adolescents and young adults with CP will be established using quantitative analysis, and the feasibility of this programme will be explored. In addition, the reliability (consistency) and measurement error (agreement) of the OMs used to evaluate the effects of an exercise programme involving adolescents and young adults with CP will be established.

1.2 Overview of the thesis

An illustration of the overview of the thesis is shown in Figure 1.1. The next chapter (**Chapter 2**) critically evaluates the available literature on the efficacy and effectiveness of exercise programmes for young people with CP through

a narrative review. Secondly, this chapter describes the OMs chosen for the evaluation of the exercise intervention, in particular with regard to their psychometric properties and feasibility/practicality. **Chapter 3** outlines the details regarding the methodology of Study 2 and Study 3, in particular the OMs used in these studies. **Chapter 4** attempts to synthesise the evidence for the psychometric properties of measures of gait quality and walking performance for young people with CP (Study 1). In this systematic review, the methodological quality of the studies assessing the psychometric properties of gait quality and walking performance in adolescents and young adults with CP will be rated using a standardised checklist (COSMIN). The strength of the evidence presented in these studies will also be assessed using standard criteria, and the overall level of evidence will be scored according to the Cochrane criteria.

Study 2, Chapter 5 aims to investigate the test-retest reliability of physical function, habitual physical activity, quality of life and self-esteem OMs in adolescents and young adults with CP and age-matched peers. **Chapter 6** will compare the OMs of physical function, habitual physical activity, quality of life and self-esteem in the young people with CP and age-matched peers who took part in Studies 2 and 3. **Study 3, Chapter 7** aims to investigate the feasibility and effects of an 18-week pragmatic community exercise programme on physical function, gait quality, habitual physical activity patterns, quality of life and self-esteem in a pilot RCT. Lastly, **Chapter 8** synthesises and discusses the outcomes and limitations of the three studies in this thesis and provides recommendations for future studies.

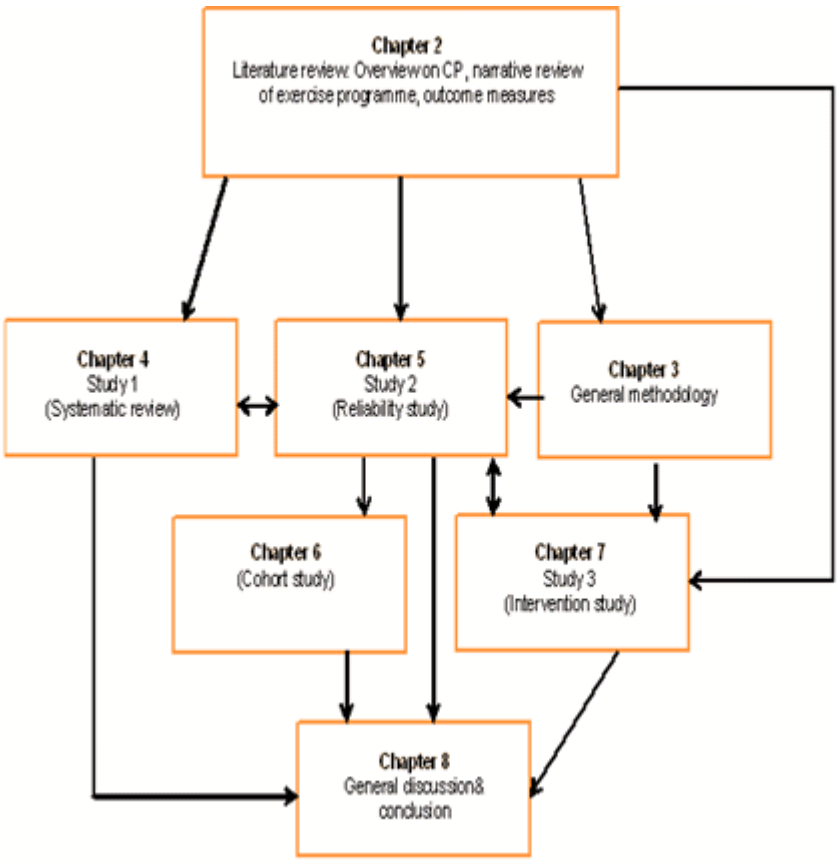


Figure 1.1 Overview of the thesis

CHAPTER 2 LITERATURE REVIEW

2.1 Introduction

This chapter will commence with an overview of CP in terms of its clinical definition, incidence, manifestation, management and therapeutic exercises for young people with CP. It will then explore the current evidence available on exercise interventions in young people with CP with a focus on exercise programmes such as aerobic training, strength training and a combination of the two. As such, this part of the literature review provides a background to the choice of the type and content of the exercise intervention evaluated as part of this thesis (Chapter 7). Next, the second part of the literature review will describe the OMs used to evaluate the effects of exercise studies and will justify the choice of the OMs used in this thesis. This literature review will then highlight the gaps identified in the evidence and explain the rationale for the studies which form part of this thesis.

2.2 Cerebral Palsy: An overview

2.2.1 Definition

Rosenbaum et al. (2006) defined CP as a 'group of permanent disorders of development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain'. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication and behaviour caused by epilepsy, and by secondary musculoskeletal problems caused by muscle contractures, bony torsion, hip displacement and spinal deformity.

2.2.2 Incidence and prevalence

CP is the most common cause of motor disability in childhood (Cans et al. 2007). A systematic review of CP incidence was conducted in 2013 and revealed the prevalence of CP to be 2.11 per 1000 live births (95% CI 1.98–2.25) worldwide (Oskoui et al. 2013). The prevalence was highest among children born prior to 28 weeks' gestation, at 111.80 per 1000 births (95% CI 69.53–179.78), whereas it was lowest, at 1.35 (95% CI 1.15–1.59), for children born after 36 weeks. In terms of birth weight, the prevalence was highest in children weighing 1000–1499 g (59.18 per 1000 live births; 95% CI 43.38–73.95) and lowest in children weighing over 2500 g (1.33 per 1000 live births; 95% CI 1.19–1.49) (Oskoui et al. 2013). In developed countries, the prevalence of CP stands at approximately 2–2.5 per 1000 live births (Odding et al. 2006, Andersen et al. 2008). In the UK, the incidence is approximately 2 in every 1000 live births, although this rises with low birth weights (Surman et al. 2009). Dependent on the subgroup of CP, 25–80% of people with CP have additional impairments such as cognitive, speech, visual and hearing (Odding et al. 2006).

2.2.3 Classification and main clinical features of cerebral palsy

In 1893, Rosenberg categorised CP as generalised rigidity, paraplegic rigidity, bilateral spastic hemiplegia, bilateral athethosis, chorioform diplegia and atypical forms. This classification was largely used for understanding and describing CP during that time (Scherzer 2001). Later, in 1950, Fay classified CP according to anatomical location and pathophysiology, and this had a strong influence on the subsequent classification by the American Academy for Cerebral Palsy (Scherzer 2001). CP has a modern-era classification that considers its early developing signs and identifies the type of motor involvement, distribution and degree of involvement and the extent of treatment required, as well as motor types (Baxter et al. 2007). The topography (which parts of the body are involved), nature of the impairments and severity are all commonly used in clinical settings when categorising

people with CP (Cans et al. 2007). Figure 2.1 displays a flow chart illustrating the classification of CP.

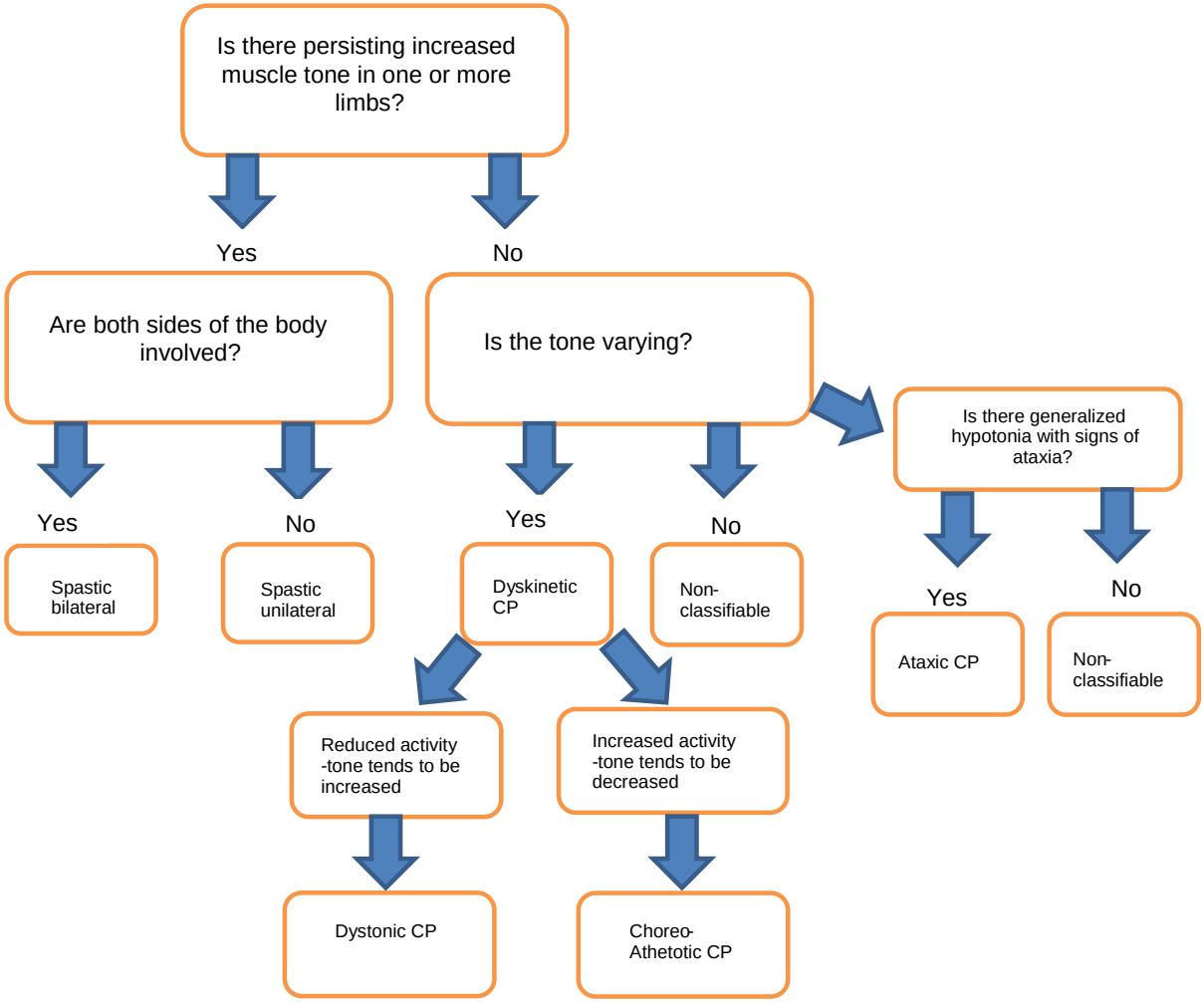


Figure 2.1 Classification of Cerebral Palsy – Adapted from the SCPE Collaborative Group

With regard to functional aspects, the Gross Motor Function Classification System (GMFCS) was introduced in the 1990s (Palisano et al. 2008a, Palisano et al. 2008b, Palisano et al. 2008c). The GMFCS provides families and clinicians with a clear description of a child's current motor function and can provide some guidance as to the type of equipment or mobility aids a child may need in the future, e.g. crutches, a walking frame or wheelchair. The GMFCS has five levels that describe the gross motor function within two age bands; 0–12 years (for children), and 12–18 years (for adolescents). The GMFCS classification for young people with CP aged 12–18 years is shown in Table 2.1.

Table 2.1 Gross Motor Classification Score (Palisano et al. 1997)

GMFCS	Description
Level I	Youth walk at home, school, outdoor and in the community. Youth are able to walk up and down curbs without physical assistance and stairs without the use of railing. Youth perform gross motor skills such as running and jumping but speed balance and coordination are limited. Youth may participate in physical activity and sports depending on personal choices and environmental factors.
Level II	Youth walk in most settings. Environmental factors (such as uneven terrain) and personal preference influence mobility choices. At school or work, youth walk using a handheld mobility device for safety. Outdoors and in the community, youth may use wheeled mobility when travelling long distances. Youth walk up and down stairs holding a railing or with physical assistance if there is no railing. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activity and sports.
Level III	Youth are capable of walking using a handheld mobility device. Compared to individuals in other levels, youth in Level III demonstrate more variability in methods of mobility depending on physical activity, environmental and personal factors. When seated, youth may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance from a person or support surface. At school, youth may self-propel a manual wheelchair or use powered mobility outdoors and in the community, youth are transported in a wheelchair or use powered mobility. Youth may walk up and down stairs holding onto a railing with supervision or physical

	assistance. Limitations in walking may necessitate adaptations to enable participation in physical activity and sports including self-propelling a manual wheelchair or powered mobility.
Level IV	Youth use wheeled mobility in most settings. Youth require adaptive seating for pelvic and trunk control. Physical assistance from 1 or 2 persons is required for transfers. Youth may support weight with their legs to assist with standing transfers. Indoors, youth may walk short distances with physical assistance, use wheeled mobility, or, when positioned, use a body support walker. Youth are physically capable of operating a powered wheelchair. When a powered wheelchair is not feasible or available, youth are transported in a manual wheelchair. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.
Level V	Youth are transported in a manual wheelchair in all settings. Youth are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and mobility but limitations are not fully compensated by equipment. Physical assistance from 1 or 2 persons or a mechanical lift is required for transfers. Youth may achieve self-mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

2.2.4 Management for people with cerebral palsy

The management of CP is not aimed at curing or achieving normalcy but rather at increasing functionality, improving capabilities and maintaining optimum health in terms of mobility, cognitive and social skills and independence (Krigger 2006). This requires a team approach that is focused on overall individual progress and not solely on the improvement of a single symptom. Treatment programmes encompass physical and behavioural therapy, pharmacological and surgical treatments, mechanical aids and the management of associated medical conditions. The goals for physical, occupational, speech and behavioural therapies include enhancing patient and caregiver interactions at the same time as providing family support. Aisen et al. (2011) suggested that the optimum team should include a neurologist, orthopaedic surgeon, psychologist, physiotherapist, occupational therapist, speech therapist and social worker. Multidisciplinary team approaches and evidence-based practice thus aim to achieve the best outcomes for individuals with CP (Aisen et al. 2011).

2.2.5 Exercise and physical activity in young people with cerebral palsy

Exercise refers to a wide range of physical activities that focus on restoring and maintaining strength, endurance, flexibility, stability and balance (Taylor et al. 2007); hence, it is important in the rehabilitation or habilitation of patients with CP (Tecklin 2008), which is recommended by health professionals such as physiotherapists. Physiotherapy is a science-based profession which helps people affected by injury, illness or disability through movement, manual therapy, education and advice, as well as exercise (The Chartered Society of Physiotherapy 2013). The aims of exercise in CP are to improve and maintain motor abilities that have been decreased through disuse, learning to adopt a healthy lifestyle and to develop appropriate community mobility (Levitt 2010).

The term ‘exercise’ is often used interchangeably with ‘physical activity’. However, there are differences in their respective definitions. Exercise is

defined as physical activity that is planned, structured, repetitive and purposive in the sense that the improvement or maintenance of one or more components of physical fitness is an objective. Physical activity is defined as any bodily movement produced by skeletal muscles that results in energy expenditure (World Health Organization 2008). However, both have a number of common elements, and in fact 'exercise' is a subcategory of physical activity (Caspersen and Merritt 1995).

The life expectancy of people with CP has increased dramatically over the course of the last two decades (Rosenbaum et al. 2002, Cooley and American Academy of Paediatrics Committee on Children with Disabilities 2004a). Studies have shown that if a child with CP lives to the age of 18, then they are more likely to live beyond the age of 40 (Hemming et al. 2005). The increasing number of adolescents and young adults with CP provides new challenges for the patients themselves, as well as for their families, health care teams and communities. Adolescence is the period in human growth and development that occurs after childhood and before adulthood, from the ages of 10 to 19. It is one of the important phases of shifts that occur during the life of an individual. It involves biological processes, such as the onset of puberty (World Health Organization 2008). Adolescence is also critical as a developmental period in that it has an impact on an individual's social integration and behaviour in adulthood. Change and adjustment may be more difficult for adolescents with a physical disability (Michelsen et al. 2006). In terms of medical needs, they will also experience the process of transition from childhood to adulthood services. Transition to adult-oriented health care, as defined by Blum et al. (1993), is a planned and purposeful movement of adolescents with chronic physical or medical conditions from child-oriented to adult-oriented health care systems and often occurs between the ages of 16 and 21 (Wright et al. 2016, Bjorquist et al. 2015). A recent study in Scotland looking at the transition from paediatric to adult health services for young people with CP showed that currently, coordination and communication within health services is lacking, in addition to between

the health services and educational, social services and adult health services to which young people are transitioning (Wright et al. 2016). Although Wright et al. (2016) did not provide any evidence that this problematic transition process was having a direct adverse effect on the health and well-being of patients, other previous studies have shown that people with a disability who are neglected between paediatric and adult health care are more likely to face high-cost emergency medical care and increasing disabilities (American Academy of Paediatrics et al. 2002).

Apart from the often problematic transition from child to adult medical services that mostly affects those between 16 and 21 years of age (Wright et al. 2015, Bjorquist et al. 2016), the evidence shows that adolescents with physical disabilities are less physically active than their peers (Carlon et al. 2013, Maher et al. 2007, Rimmer and Rowland 2008, Bloemen et al. 2015, Pitetti et al. 2013). A recent systematic review revealed that young people with CP between the ages of 5 and 18 have a lower rate (13% to 53%) of habitual physical activity than their non-disabled peers (Carlon et al. 2013). Maher et al. (2007) conducted a cross-sectional study on young people with CP aged between 11 and 17 years and found that adolescents with CP scored lower for all items in the Physical Activity Questionnaire for Adolescents in comparison with their age-matched peers; hence, this showed adolescents with CP as being less physically active than their healthy peers. Maher et al. (2007), in their study, also revealed that physical activity decreased with increasing age, as the participants with CP aged 14–17 had lower physical activity scores than the participants with CP aged 11–14 years.

Many studies have reported that people with CP show a decline in their quality of life, functional mobility, walking and overall well-being in early adulthood (Day et al. 2007, Morgan and McGinley 2014, Andersson and Mattsson 2001, Livingston and Rosenbaum 2008, Rimmer 2005). Day et al. (2007) reported the onset of a decline in ambulatory individuals with CP from the age of 25 years. This onset of functional decline may signal the start of a

downward cycle (Rimmer 2005), whereby disability, or increased disability, results in the experiencing of greater difficulty engaging in physical activity, thus leading to its avoidance. Over time, this can contribute to secondary conditions (such as pain, weight gain, reduced fitness and impaired balance) and greater functional decline, thereby exacerbating the cycle of increased disability and barriers to physical activity. The factors that contribute to this trend are beyond the scope of this report; however, some clinicians report that many individuals with CP stop exercising (i.e. physiotherapy exercise) after they have been discharged from paediatric physiotherapy services, due to them being tired of having 'physiotherapy' since childhood (Andersson and Mattsson 2001).

Physical activity in adolescents without disability may contribute to the development of healthy adult lifestyles and help in reducing chronic disease incidence in the long term (Hallal et al. 2006). The review by Hallal and colleagues also highlighted that the short-term benefits for those youth who exercise or are physically active include improvements in bone density and social interaction. A systematic review of children and adolescents (age <20) with a disability looking at the benefits of physical activity found strong evidence that activities such as group exercise programmes, treadmill training or hippotherapy have a positive impact on aerobic capacity, gross motor function and parent or participant satisfaction (Johnson 2009).

To date, there have been no physical activity guidelines that are specific to CP; however the, UK Government Department of Health has published a set of physical activity guidelines for children and adults (Davies et al. 2011), as shown in Table 2.2. These physical activity guidelines can be applied to disabled children and adult groups, including those with CP, while emphasising that these groups' physical activity (i.e. type, intensity) needs to be adjusted for each individual, based on that person's exercise capacity (fitness level) and any special health or risk issues.

Table 2.2 Guidelines for physical activity for children (5–18) and adults (19–64) (Davies et al. 2011)

Guidelines for children (5–18 years old)	Guidelines for adults (19–64 years old)
1. All children and young people should engage in moderate to vigorous intensity physical activity for at least 60 minutes and up to several hours every day. 2. Vigorous intensity activities, including those that strengthen muscle and bone, should be incorporated at least three days/week. 3. All children and young people should minimise the amount of time spent being sedentary (sitting) for extended periods.	1. Adults should aim to be active daily. Over a week, activity should add up to at least 150 mins (2½ hours) of moderate intensity activity in bouts of 10 mins or more. One way to approach this is to do 30 mins at least 5 days/week. 2. Alternatively, comparable benefits can be achieved through 75 mins of vigorous intensity activity spread across the week or a combination of moderate and vigorous intensity activity. 3. Adults should also undertake physical activity to improve muscle strength on at least two days/week. 4. All adults should minimise the amount of time spent being sedentary (sitting) for extended periods.

mins; minutes

A related behaviour, but one that is distinct from physical activity, is the amount of time spent being sedentary. Sedentary behaviour is defined as any waking behaviour characterised by an energy expenditure ≤1.5 metabolic equivalents (METs) while in a sitting or reclining posture (Barnes et al. 2012). In general, this means that any time a person is sitting or lying down, they are engaging in sedentary behaviour. Common sedentary behaviours include watching television, playing video games, using a computer and reading. Sedentary behaviour has been reported to adversely affect metabolism and cardiovascular health (Hamilton et al. 2008).

In conclusion, the evidence suggests a problematic transition process to adult health services at the same time as a decrease in physical activity around the ages of 16 to 21 and reduced mobility starting in early adulthood (around approximately 25 years of age). All of these factors can adversely impact the well-being and quality of life of young people with CP. This is very alarming and therefore proactive and sustainable management to keep adolescents and young adults with CP physically active and less engaged in sedentary behaviour is crucial.

The next section in this chapter comprises a narrative systematic review of the characteristics and results of studies investigating the effects of exercise programmes for young people (12–20 years of age) with CP.

2.3 Exercise programmes in young people with CP, a narrative systematic review of the literature

Approximately twenty years ago, strength or resistance exercise for people with CP was often avoided as it was speculated to increase their muscle spasticity. However, studies carried out in the last twenty years have shown no adverse effect on patterns of movement (Damiano et al. 1995), flexibility (Holland and Steadward 1990) or spasticity (Fowler et al. 2001) following resistance training. As a result, there has been growing interest in exercise programmes that improve lower limb muscle strength and/or aerobic capacity in people with CP. Two narrative systematic reviews (Dodd et al. 2002, Darrah et al. 1997), one systematic review (Mockford and Caulton 2008) and one meta-analysis (Park and Kim 2014) have been published reviewing the evidence regarding the effects of strength training in children with CP. Regarding aerobic training and a combination of both strength and aerobic training, the two systematic reviews have been published by Rogers et al. (2008) and Verschuren et al. (2008a), respectively (Table 2.3).

Table 2.3 Summary of systematic review and meta-analysis articles in CP exercise studies

Author	Aim	Reviewed studies		Main conclusion/ Recommendation
Dodd et al. 2002	Determine whether strength training produces beneficial outcomes for people with CP: A systematic review	Darrah et al. 1999 Damiano & Abel 1998 Toner et al. 1998 Tweedy 1997 Damiano et al. 1995	MacPhail & Kramer 1995 O'Connell & Barnhart 1995 Lockwood 1993 McCubbin & Shasby 1985 Healy 1958	Strengthening exercises for individual muscle groups increases muscle strength of young people with mild CP with no adverse effect on spasticity.
Darrah et al. 1997	Critically appraise the effects of progressive resisted muscle strengthening for children and adolescents with CP: A review	Damiano et al. 1995a Damiano et al. 1995b MacPhail and Kramer 1995 Holland and Steadward 1990	Horvat 1987 McCubbin and Shasby 1985 Healy 1958	The relationship between strength training and functional abilities is scarce.
Park and Kim 2014	Synthesise the effects of strengthening on individuals with CP : A meta-analysis	Scholtes et al. 2012 Chen et al. 2012 Fowler et al. 2010 Maeland et al. 2009 Unnithan et al. 2007 Lee et al. 2007	Unger et al. 2006 Kerr et al. 2006 Engsberg et al. 2006 Van der Linden et al. 2003 Doss et al. 2003	Optimal duration of exercise of 40 to 50 minutes per session performed three times per week for increasing muscle strength in people with CP.
Mockford and Caulton 2008	Determine the evidence regarding progressive strength training for children and adolescents with CP who are ambulatory: A systematic review	Liao et al. 2007 Ensberg et al. 2006 Jiang et al. 2006 Unger et al. 2006 Morton et al. 2005 Eagleton et al. 2004 Dodd et al. 2003	Blundell et al. 2003 Damiano and Abel 1998 Tweedy 1997 Damiano et al. 1995a Damiano et al. 1995b MacPhail & Kramer 1995	Function and gait improvement were greater in pre-adolescents in comparison to adolescents.
Rogers et al. 2008	Evaluate the evidence for the effectiveness of aerobic exercise intervention for children with CP	Fragala-Pinkham et al. 2005 Schlough et al. 2005 Mulligan et al. 2004 Shinohara et al. 2002 Van den Berg-Emons et al. 1998 Wiepert & Lewis 1998 Darrah et al. 1998	Rintala et al. 1990 Dresen et al. 1985 Bar-Or et al. 1976 Berg 1970 Lunderberg & Pernow 1970 Lunderberg et al. 1967	More research with rigorous methods is required to determine a specific set of exercise guidelines and safety considerations.

Author	Aim	Reviewed studies		Main conclusion/ Recommendation
Verschuren et al. 2008a	Investigate the effects of all types of exercise programmes focusing on cardiovascular fitness (aerobic and aerobic capacity) and/or lower-extremity muscle strength in children with CP: A systematic review	Patikas et al. 2006 Unger et al. 2006 Morton et al. 2005 Schlough et al. 2005 Eagleton et al. 2004 Dodd et al. 2004 Dodd et al. 2003 McBurney et al. 2003 Blundell et al. 2003 Shinohara et al. 2002	Damiano et al. 1998 Van den Berg-Emons et al. 1998 Rintala et al. 1998 Hutzler et al. 1998 Darrah et al. 1997 Damiano et al. 1995 MacPhail &Kramer 1995 Berg et al. 1970 Lunderberg et al. 1967 Healy 1958	More studies are warranted to investigate the efficacy of exercise programmes on daily activity, participation level and quality of life in children with CP.

The aims of exercise programmes designed recently for people with CP are not just to prevent secondary complications and/or impairments, i.e. lack of range of motion, muscle weakness and spasticity, from getting worse (Damiano et al. 2002) but also to maximise the individual's overall health (Katsimanis et al. 2002, Damiano and Abel 1998) and quality of life (Groff et al. 2009). According to the World Health Organization (2008), health is regarded as a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity.

A recent cross-sectional study of 70 young people (11–17 years) with CP (GMFCS levels I–V) revealed that the level of physical activity significantly predicted their health-related quality of life (R^2=0.64, p<0.01), social quality of life (R^2=0.28, p<0.01) and happiness (R^2=0.08, p<0.01). The authors concluded that there is a need for clinical services and intervention studies aimed specifically at increasing physical activity in young people with CP (Maher et al. 2007).

2.3.1 Methods: Identification of studies in the literature review

A systematic search of the literature was performed to identify the exercise studies discussed in this literature review using the online databases MEDLINE, CINAHL, PubMed and Scopus up to December 2016. Search terms included subject headings and text words based on (I) cerebral palsy; and (II) exercise, strength, resistance, fitness, working capacity, aerobic power, anaerobic power, endurance and cardiorespiratory physical training. In this review, the term young people is used in reference to adolescents, and the term young adults refers to those between 12 and 20 years of age (World Health Organisation 2008).

The studies had to meet the following inclusion criteria to be included in this review: 1) any experimental, pre-post and case studies involving at least one participant aged 12–20 with CP; 2) any land-based exercise intervention aimed at strengthening lower-limb muscles and/ or improved aerobic capacity; 3) any measure of strength, body function, activity, participation, self-esteem and quality of life; and 4) full-text original articles in English.

2.3.2 Results

The search strategy resulted in the identification of 272 articles. Initial screening based on the tittle resulted in the exclusion of 166 articles. A further 42 and 35 articles were omitted based on the review of abstracts and full text, respectively. Based on the inclusion criteria regarding age, a further ten articles were excluded. Three articles were added as a result of scanning the reference lists of the primary studies. This gave a final number of 30 articles published between 1958 and 2016. The types of exercise programmes investigated are categorised as follows: strength training (n=16), aerobic training (n=4) and combined training (n=5). The details of the studies are shown in three different tables: exercise interventions focusing on strength training (Table 2.4), aerobic training (Table 2.5) and those incorporating a combination of both strength and aerobic training (Table 2.6).

The specific results obtained for each of these types of exercise training are described separately in the next three sections with regard to the demographics of the participants, the duration of the exercise and the results of the studies, followed by a summary.

2.3.2.1 Strength training

a. Demographics of the participants, study design and intervention characteristics

The number of participants included in the studies ranged from 7 to 49, with only four studies with a sample size of more than 30 participants (Patikas et al. 2006, Unger et al. 2006, Taylor et al. 2013, Scholtes et al. 2010). Given that the sample size tends to be rather small in CP studies, especially in RCTs, multi-centre trials would be an alternative to overcome the small sample size.

The age of the participants ranged from 6 to 22 years. Nine studies described their participants' GMFCS level (Dodd et al. 2003, McNee et al. 2009, Eek et al. 2008, Bania et al. 2016, Taylor et al. 2013, Scholtes et al. 2010, Chen et al. 2012, McBurney et al. 2003, Auld and Johnston 2014). Dodd et al. (2003) included a majority of participants who were classified as GMFCS level III (n=9). The majority of the participants in the study by Taylor et al. (2013) were classified as GMFCS II. Overall, the majority of the participants comprised those less severely affected, with no studies including people with CP classified as GMFCS levels IV–V. Hence, identifying the effect of strength training across the GMFCS levels is essential.

Ten studies conducted pre- and post-test design with no control group, with the remainder (n=9) being RCTs, none of which adopted a double-blind design. Double-blinding is where neither the participants nor the assessors are aware of the experimental groups until after the data have been collected (Portney and Watkins 2000). To blind participants, the control group should be offered a placebo. However, for many rehabilitation research studies, this

is not always possible; hence, a single-blind design may be employed instead, whereby only the assessor is blind. Only two studies (Taylor et al. 2013, Scholtes et al. 2010) used a single-blind design. Blinding will substantially strengthen the study as it will reduce the risk of bias when assessing the participants.

The type of strength training varies throughout the literature, with many studies using dumb-bells, backpacks, body weight or TheraBands/resistance bands; further details can be seen in Table 2.2. Six of the exercise programmes (n=6) in this review were home-based training (Dodd et al. 2003, Damiano et al. 1995, Damiano and Abel 1998, Chen et al. 2012, McBurney et al. 2003, Dodd et al. 2004). Four studies (n=4) utilised the community gymnasium (Taylor et al. 2013, Auld and Johnston 2014, Tweedy 1997, Eagleton et al. 2004). Other studies were conducted at special schools (n=2) (Unger et al. 2006, Scholtes et al. 2010), hospital (n=1) (Patikas et al. 2006) and a university laboratory (n=1) (MacPhail and Kramer 1995). The remaining studies were carried out at both home and hospital (McNee et al. 2009, Eek et al. 2004). Whilst the implementation of a programme at a community gymnasium might not always be possible, this type of venue does have the benefit of providing a conducive, welcoming and inclusive community environment. It has also been shown that a community setting is an important element that contributes to the success of an exercise programme in adults with CP (Allen et al. 2004).

The duration of exercise programmes reported in the literature ranges from 6 weeks to 40 weeks. The majority of the studies conducted programmes for 6 (n=5) and 8 weeks (n=5), with the remainder being for durations of 10 weeks (n=2) and 12 weeks (n=4). It was recommended that longer interventions with progressive intensities (i.e. 12–16 weeks) were essential in order to experience any significant strength improvement in people with CP (Verschuren et al. 2008a). The frequencies of exercise per week varied across the studies, with the majority (n=10) implementing twice- or thrice-

weekly sessions, and these were conducted in accordance with the guidelines stated by (American College of Sports Medicine 1980, Faigenbaum et al. 2009).

b. Results and discussion of the studies

A randomised clinical trial evaluated the effects of a home-based, six-week strength-training programme aimed at increasing the lower limb strength of 21 people with CP aged between 8 and 18 years (GMFCS I–III) (Dodd et al. 2003). There were significant improvements in the strength of the ankle plantarflexors and knee extensors of the experimental group compared to the control group from baseline to week six. No significant improvements were found in terms of the ICF 'activity' domain as measured by GMFM dimension E over the first six weeks between the experimental and control group. However, the authors noted that there was a trend of improvement in the experimental group compared to the control group, and power analysis revealed that if the effect size were maintained and the sample size increased to n=26 in each group, there was an 80% chance that the comparison would reach statistical significance. The levels of participation and adherence to the training programme were reported to be excellent; however, none of the participants continued to exercise after the study had ended. The study did not provide information on whether the participants did any other form of exercise or physical activity after the end of the programme. Thus, the study in question reveals that although there was excellent adherence to the home-based exercise programme during the course of the study, the intervention did not prove sustainable after its completion.

A previous RCT investigating the effect of eight weeks' resistance training (one to three times per week) on young people with CP aged 13–18 years found no significant improvement with regard to gait velocity, cadence and stride length. In addition, Unger et al. (2006) reported that no significant improvement was found in the participants' kinematics following the

programme (Unger et al. 2006). Interestingly, there was a significant difference in terms of the participants' perception of their ability to walk between the classroom and managing stairs without support.

A gymnasium-based strength-training programme was conducted by Taylor et al. (2013). This trial by Taylor et al. (2013) involved 48 participants aged between 14 and 22 years. The participants performed 12 weeks' progressive resistance training twice weekly and showed an increment in muscle strength. However, this effect did not carry over into any objectively measured improvements in mobility, such as the six-minute walk test (6MWT), gait kinematics and GMFM. These authors also emphasised the importance of the safety and feasibility of any exercise programme aimed at helping young people with CP become stronger and achieve physical activity guidelines (Taylor et al. 2013).

A study comprising 17 participants with CP aged 12 to 20 years revealed that there was a statistically significant increase in the peak torque (Nm/kg) of knee extensor and flexor muscles, as well as of the GMFM score following strength training three times per week for a total of eight weeks (MacPhail and Kramer 1995). However, the study's authors did not find any significant changes in spasticity and gait parameters such as walking velocity and energy efficiency during walking.

In contrast to previous results (MacPhail and Kramer 1995, Unger et al. 2006), Eagleton et al. (2004) found statistically significant improvements in spatial-temporal gait parameters (step length, velocity, cadence and energy expenditure index) following strength training conducted three times per week for six weeks involving seven young people with CP aged 12 to 20 years. Of note, this study was the first strength-training programme to be conducted in a community gymnasium since strength training for young people with CP was first reported by Healy in 1958. Eagleton et al. (2004) acknowledged that all the participants reported that they enjoyed the training

in the gymnasium. Safety is a vitally important element for a successful exercise programme as it contributes to a further increase in the confidence of the participant to continue the programme once the study is finished. Ploughman et al. (2014) suggested that people with disabilities need education, professional support, appropriate programming and accessible space to participate in a community-based exercise programme. People with disabilities often feel insecure if the exercise programmes are not specially designed to meet their needs or they lack support from physiotherapists and/or fitness instructors (Lennon et al. 2013, Simpson et al. 2011).

Early studies (Healy 1958, Horvat 1987) showed the positive effects of strength training. The strength training conducted by Healy (1958) compared isometric (static contraction) and isotonic (dynamic resistance) techniques in five participants aged 8 to 16 who exercised three times per week for eight weeks and who showed improvement in knee extensor strength at an eight-week assessment. The study also found improvement in the range of motion of knee extension but no significant difference between the isometric and isotonic techniques. A single case study involving a 21-year-old participant with spastic hemiplegia also demonstrated an improvement in knee extensor strength after eight weeks of strength training (Hovart 1987). Neither study (Healy 1958, Horvat 1987) reported other outcomes, however, such as walking speed and gross motor function.

The first systematic review by Darrah et al. (1997) identified six strength exercise studies. Later, Dodd et al. (2002) carried out a systematic review and included the same studies as those reviewed by Darrah et al. (1997), with the inclusion of an additional five strength exercise studies. Both reviews revealed that strengthening exercises for individual muscle groups increased the muscle strength of young people with mild CP with no adverse effect on spasticity. However, evidence pertaining to the relationship between improved muscle strength and improved functional ability is scarce. The third systematic review by Mockford et al. (2008) on progressive strength training

in children and youths with CP who are ambulatory suggested such training may improve motor development in younger children; however, this may not be the case in youth as deterioration may occur. Mockford et al. (2008) acknowledged that improvement in some aspects of gait (i.e. cadence, walking speed) may occur. A recent meta-analysis by Park et al. (2014) on the effect of strengthening interventions in individuals with CP identified 13 RCT studies published between 2001 and 2012. The results of this meta-analysis suggested an optimal exercise duration of 40 to 50 minutes per session performed three times per week for increasing muscle strength in people with CP. Previous systematic reviews and meta-analyses recommended higher-quality studies (i.e. RCT) to investigate the effects of strength training in relation to mobility (Park et al. 2014, Darrah et al. 1997, Dodd et al. 2002), function (Darrah et al. 1997, Dodd et al. 2002) and participation (Darrah et al. 1997, Dodd et al. 2002).

c. Summary of evidence on the effects of strength training

As discussed earlier, strength training can result in an improvement in muscle strength (MacPhail and Kramer 1995, Taylor et al. 2013, McNee et al. 2009, Eek et al. 2008, Chen et al. 2012, Scholtes et al. 2010, Auld et al. 2014, Damiano et al. 1995, Damiano et al. 1998, Tweedy et al. 1997), gross motor function (MacPhail and Kramer 1995, Dodd et al. 2003, Patikas et al. 2006, Eek et al. 2008, Damiano et al. 1998), gait parameters (Eagleton et al. 2004, Eek et al. 2008, Damiano et al. 1998) and quality of life (McBurney et al. 2003). However, there is a need for further exploration in this area, as discussed in section 2.3.3.

2.3.2.2 Aerobic training

a. Demographics of the participants, study design and intervention characteristics

The number of participants across the studies ranged from 3 to 22. The age of the participants ranged from 6 to 25 years. GMFCS was reported in only

one study, with one participant classified as GMFCS I and two as GMFCS III (Schlough et al. 2005).

The majority (n=3) of the studies were pre-posttest design with no control group, and one study was a case series with three participants. The type of exercise varied, with two studies utilising a cycle ergometer (Shinohara et al. 2002, Berg 1970), one study using several different modalities, i.e. treadmill, elliptical and recumbent stepper (Schlough et al. 2005), and the remaining study (n=1) including activities such as running and jumping (i.e. Lundberg et al. 1967) to achieve the desired aerobic effect in their participants. In terms of the venue of the programmes, community gymnasiums and university laboratories were utilised for the training.

The duration of the exercise programmes in all of the studies was between 6 and 20 weeks. The frequencies with which they were carried out were between two or three times per week, for a duration of 20 to 30 minutes per session.

b. Results and discussion of the studies

Lunderberg (1967) and Berg (1970), in their respective studies, found that participants with CP had improved their aerobic capacity at the end of the training. Lunderberg (1967), in a study of 14 participants with CP aged 15–20 years, reported increases in the participants' maximal oxygen uptake (VO_2max) after aerobic training (i.e. running, jumping) twice a week for a period of six weeks. Similarly, Berg (1970) reported improvement in the 22 participants with CP aged 12 to 25 years who carried out exercise with a bicycle ergometer three times per week for between 1.5 and 6 months. The VO_2max of the participants was reported to increase by between 10 and 25 per cent following the total training periods that varied between 1.5 and 16 months (Berg 1970). Whilst early studies of aerobic training (Lunderberg 1967, Berg 1970) showed improvement in the participants' VO_2max following the training, both studies reported only the pre- and post-intervention means;

however, they provided no evidence in terms of statistical analysis to support their claims and the results should thus be interpreted with caution.

Shinohara et al. (2002) investigated 11 participants with CP aged between 13 and 16 years who were randomly divided into two groups. Six of the 11 participants exercised using a cycle ergometer, with the other five participants carrying out training using an arm cranking ergometer twice a week. They found that the VO_2max of the adolescents who exercised on a cycle ergometer showed significant improvement in comparison to the group that trained with the arm cranking ergometer, for which no change in VO_2max was reported. The training period for both groups ranged from 6 to 20 weeks. Both the study by Shinohara et al. (2002) and previous studies (Lunderberg 1967, Berg 1970) appeared to examine only one outcome. In contrast, in a report of three case studies, Schlough (2005) evaluated the outcomes of aerobic exercise on participants with CP aged between 17 and 20 years across a much wider range of aspects, i.e. energy expenditure index (EEI), strength, GMFM and self-perception. The participants exercised three times per week for six weeks using an elliptical trainer, treadmill or recumbent stepper. This study revealed a significant improvement in the participants' EEI, muscle strength and velocity in the individual cases (participants) following the programme.

Rogers et al. (2008) undertook a systematic review that aimed to evaluate the effect of aerobic exercise for children with CP. The review included land-based and non-land-based (i.e. hydrotherapy) exercise programmes. The authors concluded that more research involving rigorous methods was required to determine a specific set of exercise guidelines and safety considerations.

c. Summary of evidence on the effects of aerobic training

A number of researchers have examined the effects of aerobic training in young people with CP. Of four studies (Lunderberg 1967, Berg 1970,

Shinohara et al. 2002, Schlough 2005), the majority (n=3) examined the effects of VO_2max. It was shown that, in general, aerobic training benefits aerobic capacity. Only one study (Schlough 2005) investigated other aspects and reported improvements in strength, walking efficiency, gross motor function and self-perception. However, this study appeared to employ poor methodologies (i.e. a very low number of participants, non-RCT). There is a need for further research in this area, and this will be discussed in section 2.3.3.

2.3.2.3 Combined training

a. Demographics of the participants, study design and intervention characteristics

The number of participants included in the studies ranged from 13 to 65, with only two studies recruiting less than 30 participants. The age of the participants ranged from 7 to 24 years. Four out of the five studies reported the GMFCS level of the participants in the individual studies. Three of these four studies reporting GMFCS levels had the largest number of participants with CP classified as GMFCS level I, followed by participants with CP classified as GMFCS level II.

Four of the studies were RCTs with the assessors being blind to the participants' group allocation. Only one study adopted a pre-test/post-test design. The venues used in the combined training studies were either community gymnasiums or rehabilitation centres.

The durations of the exercise programmes carried out were between 10 and 32 weeks. Similar to the strength and aerobic studies, the frequency of combined training was either two or three times per week.

b. Results and discussion of the studies

A previous RCT aimed at exploring the effectiveness of a 12-week exercise programme on submaximal exercise intensity during arm cranking without

resistance at 4 minutes, VO_2max and gross motor function (Unnithan et al. 2007). Thirteen participants with CP aged between 14 and 18 years took part in this programme. The experimental group took part in 70 minutes' training that consisted of a warm-up, 20 minutes' strength training and 20 minutes' aerobic training and stretching, three times per week for 12 weeks. The control group continued with their normal physiotherapy session. The results showed that submaximal exercise intensity and aerobic capacity were significantly improved in the exercise group relative to the control group at week 12. They also reported a significant increase in GMFM scores (dimensions D and E) in the exercise group compared to the control group. This study showed that a combined strengthening and aerobic training programme of 12 weeks' duration had the ability to positively impact on the participants' fitness and gross motor function.

Another RCT was conducted with 68 participants with CP aged between 16 and 24 years (Verschuren et al. 2007). The participants in the exercise group followed a programme that consisted of functional aerobic, anaerobic training and muscle strengthening twice weekly for a period of eight months, in addition to their usual care. The participants in the control group received their usual rehabilitation care. The results of the study revealed that there were significant improvements in aerobic capacity measured by the 10 m shuttle run test and anaerobic capacity measured using the Muscle Power Sprint Test at 4-month and 8-month assessments in the experimental group. Moreover, the participants' muscle strength of lower extremities was found to have increased by 20 per cent in comparison to the control group at 8 months. The number of participants in the study was higher compared with the previous RCT study (i.e. Unnithan et al. 2007) as the participants were recruited from different centres.

Fowler et al. (2010) conducted a multi-centre RCT that investigated the effects of a cycling intervention on 55 participants with CP aged between 7 and 18 years. The cycling intervention known as PEDALS was performed in

a community gymnasium three times per week for a duration of 12 weeks. It utilised stationary bicycles which provided resistance. The results showed statistically significant improvements in the 600-Yard Walk-Run Test and GMFM 66 (dimensions D & E) scores, knee extensor moments (Nm/Kg) at 120^0/s and knee flexor moments (Nm/Kg) at 30^0/s in the exercise group following 12 weeks' training compared to the baseline. However, this study found no significant differences between the exercise and control groups.

A Dutch multi-centre RCT known as LEARN2MOVE was conducted aimed at increasing physical activity and improving the physical fitness of people with CP aged between 16 and 24 years (Slaman et al. 2010). The LEARN2MOVE programme consisted of the following three parts: 1) counselling on daily physical activity by a physiotherapist once a month for 6 months; 2) physical fitness training that included aerobic and strength training for 24 weeks (12 weekly supervised sessions in the gymnasium plus 12 sessions at home); and 3) counselling on sports participation for approximately 2 to 4 sessions, each of 30 minutes' duration. The results showed a statistically significant difference in VO_2max at anaerobic threshold between the exercise and control groups. The programme was effective in decreasing fatigue (p=0.02) and increasing quality of life with regard to bodily pain (p=0.01) and mental health (p=0.03) assessed during follow-up (Slaman et al. 2014a). However, no significant difference was reported in the strength of hip flexors, abductors and knee extensors between the groups following the exercise programme (Slaman et al. 2014b) nor in the objectively measured outcomes of physical activity or sedentary behaviour (Slaman et al. 2015).

Darrah et al. (1999) conducted a community-based exercise programme combining aerobic, flexibility and strength training for adolescents with 23 participants with CP aged 11–20 years. The participants trained three times per week for 10 weeks. According to Darrah et al. (1999), the training frequency and duration of the programme was based on recommendations as outlined by the American College of Sports Medicine (1991). The training,

inclusive of a warm-up, consisted of progressive aerobic training (10–30 minutes), progressive strengthening of upper-limb and lower-limb muscles and stretching. The results showed statistically significant improvements in the strength of the participants' knee extensors, hip extensors and hip abductors post training at the 10-week follow-up. However, no significant changes were found in EEI and submaximal heart rate. The authors commented that a possible reason for the lack of changes in the cardiorespiratory endurance of the participants could be related to the relatively short duration of the exercise programme (i.e. 10 weeks).

c. Summary of evidence on the effects of combined training

Combined or mixed training studies enable participants to benefit from strength, aerobic and other forms of exercise i.e. stretching. Previous combined exercise studies have reported improvements in aerobic capacity (Verschuren et al. 2007), anaerobic capacity (Unnithan et al. 2007, Verschuren et al. 2007, Slaman et al. 2014a), muscle strength (Darrah et al. 1999, Fowler et al. 2010, Verschuren et al. 2007), self-perception (Verschuren et al. 2007), gross motor function (Unnithan et al. 2007, Fowler et al 2010), fatigue (Slaman et al. 2014a) and quality of life (Demuth et al. 2012, Slaman et al. 2014a). However, further investigation is warranted to strengthen the present evidence, and this will be outlined in the next section.

Table 2.4 Studies of strength training in young people (12–20 years) with cerebral palsy

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
MacPhail and Kramer (1995)	Pre-post test	n=17, 12–20 years (mean 15.8 years), GMFCS-not specified	1) Before 2) After 3) 3-month f/up, 8 weeks' strength training, 3x/week, supervision: not specified, research laboratory, training programme: 3 sets of 5 max effort at 90%; knee flexors and extensors	1) Isokinetic (dynamometer) 2) GMFM D & E 3) EEI at free and fast walk speed, 4) Velocity over 3 mins, free and fast speeds 5) MAS	1) Peak torque increased 25% at 8w, 17% at f/up. Work increased 21% at 8w, 15% at f/up ($p<0.001$) 2) 9 participants increased scores ($p=0.011$), no change in 7 participants, decreased in one participant 3) no sig change 4) no sig change 5) no sig change
Dodd et al. (2003)	RCT	n=21, 8–18 years (mean 13.1),GMFCS (I=7, II =5, III=9)	1) Start 2) 6w 3) 18w f/up, 6-week strength training, 3x/week, supervision: physio & parent, home, training programme: 3 sets of 8–10 reps of ankle plantar flexor, knee extensor, hip extensor, exercises loaded with backpack, weight increased gradually, Logbook to record weight used (backpack), number of sets and repetition, Control group: continue normal routines such as sports, school and physiotherapy session	1) Isometric (handheld dynamometer) 2) GMFM D&E 3) Timed stair test 4) Self-selected walk speed	1) sig increased in combined knee ext &PFat 6w & 18w ($p<0.05$) 2) non-sig change 3) non-sig change 4) non-sig change

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Dodd et al. (2004)	RCT	n=17, 8–16 years (mean 12.1),GMFCS (I=6, II-4, III-7)	As above	1) Self-concept (Self-Perception Profile for Children)	1) Sig improve in scholastic competence and social acceptance ($p<0.05$)
Eagleton et al. (2004)	Pre-post test	n=7, 12–20 years (mean not specified), GMFCS level not specified	1) Before 2) After, 6 weeks' strength training, 3x/week, physio, local & school gym, training programme: 80% of 1RM of trunk and lower limbs muscle	1) Isometric (handheld dynamometer) 2) Distance walked in 3 mins 3) Visual gait analysis 4) EEI (HR before & after 2-min walk	1) No result (due to equipment failure) 2) sig increase in 6 of 7 participants ($p=0.05$) 3) sig improvement in velocity, step length, cadence ($p=0.05$) 4) sig decrease in 4 of 7 participants ($p=0.05$)

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Unger et al. (2006)	RCT single-blind	n=31, 13–18 years (mean 16.1),GMFCS-not specified	1) Before 2) After 3) 4w f/up, 8-week strength training,1-3x/week, research assistant, special school, training programme: 1–3 set of reps, 8–12 individual selection of a 28-station circuit upper and lower limb, trunk, exercise inclusive of a 5-min warm-up and stationary bicycle; Exercise progressively increased as outlined by McArdle et al. (1996); Equipment used was dumbbells, ankle and wrist cuff weights and bar-with disc weights, elastic-rubber band and gymball	1) 3DGA 2) Self-perception questionnaire	1) sig improved sum of hip/knee/ankle angles at midstance for exs group compared to controls (p=0.05).no sig change in gait parameters 2) sig improvement in perception of body image (p=0.01)
Patikas et al. (2006)	RCT	n=39, 6–16 years (mean 10.1 years), GMFCS not specified	1) presurgery and pretraining 2) 1-yr postsurgery 3) f/up, 8 weeks' strength training, 1–3x/week, physio & parent, hospital, training programme: 2 sets of 5 reps 7 exercises-hip, knee and ankle extensor and flexors	1) 3DGA 2) GMFM 3) EEI 4) MAS	1) no sig difference between group in spatiotemporal, kinematics & kinetics 2) Sig improved dimension D 1yr postsurgery in exs group only (p<0.05), no sig difference between group 3) Sig improved in EEI 1yr postsurgery in exs group only (P<0.05), no sig difference between group 4) No sig change

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Taylor et al. (2013)	RCT Single-blind	n=48, 14–22 years (18.2), GMFCS (II=29,III=19)	1) Before 2) After 3) 24w f/up, 12 weeks' strength training, 2x/week, physio, community gym, training programme: 3 sets of 10–12 reps, exs based on instrumented gait analysis and targeted muscles contributing to walking difficulties, Control group: continued usual physiotherapy and usual recreation activity	1) 6MWT 2) GMFM D & E 3) GPS 4) FMS 5) FAQ 6) Isometric strength (handheld dynamometer)	1) No sig change 2) No sig change 3) No sig change 4) Sig change at 12w between groups ($p<0.05$) 5) Sig change at 12w between groups ($p<0.05$) 6) Sig change at 12w between groups ($p<0.05$)
Bania et al. (2016)	As above	As above	As above	1) Objective measurement of Physical activity (ActivPAL^{TM})	1) No significant change
McNee et al. (2009)	Pre-post test	n= 13, 6–16 years (10.11), GMFCS (I=6,II=5,III=2)	1) Before 2) 5w 3) After 4) 12w f/up, 10 weeks' strength training, 4x/week, physio, home & tertiary centre, training programme: 3/4 sets of 6–12 reps isotonic PF using TheraBand or body weight or back pack depending on individual strength, exercise logbook to record the training	1) Passive range of motion 2) Muscle volume (3D tracking system) 3) TUG 4) FMS 5) FAQ 6) number of heel raises	1) No sig change 2) Sig improvement at weeks 5, 10 and f/up 3) No sig change 4) No sig change 5) No sig change 6) Sig improvement at weeks 5, 10 and f/up

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Eek et al. (2008)	Pre-post test	n=16, 9–15 years (9.4),GMFCS (I=10,II=6)	1) Before 2) After 8-week strength training, 3x/week, physio & parent, physio department & home, training programme: strengthening of hip ext/flex/abd/add, knee ext/flex, ankle DF/PF	1) Isometric strength (dynamometer) 2) GMFM D&E 3) 3DGA 4) ROM 5) MAS	1) Sig increase in targeted muscle group strength (hip ext, hip add, hip abd, PF) $p<0.05$ 2) Sig change $p<0.05$ 3) Sig change in cadence $p<0.05$ 4) Sig change in ROM of knee ext $p<0.05$ 5) No sig change
Chen et al. (2012)	RCT	n=28, 6–12 years (8.7), GMFCS (I=22, II n=6)	1) Before 2) After 12 weeks' virtual cycling training, 3x/week, research assistant, home-based, training programme: 20-minute cycling using stationary cycle-progressive increase resistance, Control group – continue usual physical activity	1) Bruininks-Oseretsky Test of Motor Proficiency 2) Isokinetic Strength (isokinetic dynamometer)	1) No sig change 2) Sig change in Knee ext/flexor, $p<0.05$

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Scholtes et al. (2010)	RCT Single-blind	n=49 6 to 13 years (10.4 years) GMFCS (level I n=25, level II n=17, level III n=7)	1) Before 2) During training 3) immediately after training 4) 6-week follow-up, 3x/week for 12 weeks, research assistants, special schools, training programme: 5–10mins warm-up (stretching and aerobic), leg press, sit-to-stand, lateral step up, half knee-rise with weight, lateral step up with body weight, 5–10mins cool down (stretching), control group – continue physiotherapy programme	1) Mobility as measured by GMFM 66, MobQues28, sit-to-stand test and lateral step up test 2) isometric strength (handheld dynamometer) 3) spasticity	1) No sig 2) Significant improvement for knee extensor, hip adductor 3) No sig
Auld et al. (2014)	Pre-post test	n=10, 8–15 years (mean not specified), GMFCS (I=6, II=4)	1) Before 2) After 8 weeks' strength & balance training, 1x/week, physio, community gym, training programme: 2–3 sets of 10 reps, Upper limb and lower limb strength, core stability, balance training	1) Isometric muscle strength (dynamometry), 2) Functional muscle strength (seated throw, distance jump, vertical jump, lateral step up) 3) Balance	1) Sig change in elbow flex, hip abductor, ankle dorsiflex, plantarflexor of the dominant side ($p<0.05$) 2) Sig change in seated thrown distance jump 3) Sig change in lateral reach, forward reach, lateral step
Damiano et al. (1995)	Pre-post test	n=14, 6-14 years (9.1), GMFCS: not specified	1) Before 2) 3w 3) 6w, 6 weeks' strength training, 3x/week, not described, home-based, training programme: 4 sets of 5 reps, Isotonic open-chain concentric/eccentric knee extension-4/5 reps, loads 65% of max	1) isometric of knee extensor and flexor (handheld dynamometer)	1) Significant increased quadriceps at 30, 60, 90 degrees ($p<0.001$)

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Damiano et al. (1995)	As above	As above	As above	1) 3DGA	1) No sig change
Damiano et al. (1998)	Pre-post test	n=12, 6–12 years (8.8), GMFCS not specified	1) Before 2) 6w 3) 18w f/up, 6 weeks' strength training, 3x/week, physio & parent, home-based, 3 sets of 8–10 reps, Isotonic open-chain exercise of lower limb	1) Isometric strength of hip flex/ext, abd/add; knee ext/flex; ankle flex/ext 2) GMFM 3) 3DGA 4) EEI	1) Sig increased in target muscle ($p<0.05$) 2) Sig improved Dimension E ($p<0.05$) 3) Sig improvement in velocity, cadence and double support ($p<0.05$) 4) No sig change
Engsberg et al. (2006)	RCT	n=15 6–13 years (9.5 years); GMFCS not specified	1) Before 2) After 12 weeks' strength training, 3x/week, physio, physio gym, training programme: 3 sets of 5 reps, Isokinetic exs, concentric and eccentric 3 sets/5 reps, Group 1:DF, group 2:PF, group 3: DF&PF Control (group 4): continue daily activity	1) Isokinetic strength ankle DF/PF(isokinetic machine) 2) Spasticity ankle PF 3) Passive ROM 4) GMFM 5) 3DGA 6) PedsQoL	1) Sig increase in trained eccentric muscle at 30, 90 degrees ($p<0.05$) 2) Sig reduced spasticity in exs group $p<0.05$ 3) No sig change 4) Sig increase for dimension E in exs group $p<0.05$ 5) Sig improvement in knee flexion minimum $p<0.05$ 6) Sig improvement in parent report, $p<0.05$

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Tweedy et al. (1997)	Pre-post test	n=12,10–18 years (13.8), GMFCS not specified	1) Before 2) After 10 weeks' strength training, 3x/week, supervision, gymnasium, training programme: Isotonic closed and or open-chain concentric/eccentric hip and knee extensors-weighted boots, weight machines, training diary	1) Isometric quads strength at 80-, 65-, 50-, 35-, 20-degree extension 2) Isokinetic strength quads at 60 degrees 3) Flexibility-passive knee flex/ext, active knee ext 4) Muscle tone of the knee-isokinetic movement at 60, 40, 20, 12 degrees	1) Sig improvement in knee extension at 80, 65, 50 degrees; p<0.05 2) Sig increase in peak torque knee extension; p<0.01 3) Sig ROM knee flexion (p=0.005) 4) Significant decrease right leg resistance to passive movement; p=0.046
McBurney et al. (2003)	Pre-post test	n=11,8–17 years (12.9), GMFCS (I=2, II=2, III=7)	1) After 6 weeks' strength training, 3x/week physio & parent, home-based training programme: 3 sets of 8–10 reps, bilateral half squats, heel raises, and step-ups: Ankle plantarflexor, knee extension, hip extension	1) In-depth interview on body function, activity, participation (ICF model)	2) Improvement in perception of strength, flexibility, posture, walking and the ability to negotiate stairs, improvement in mobility, improvement in school, leisure and family events

3DGA; three-dimensional gait analysis, abd; abduction,add; adduction, DF; dorsiflexion, EEI; energy expenditure index, exs; exercise, ext; extension, FAQ; Gillette Functional Assessment Questionnaire, flex; flexion, FMS; functional mobility scale, f/up; follow up, physio; physiotherapy, GMFCS; Gross Motor Functional Classification System, GMFM; Gross Motor Function Measure, dimension D; standing, dimension E; running,jumping, walking ICF; International Classification of Functioning, disability and health, MAS; Modified Ashworth Scale, max; maximum, mins; minutes, MobQues28; 28-item version mobility questionnaire for ambulant children with Cerebral Palsy,PedsQoL; Pediatric Quality of Life Inventory, PF; plantarfexion,RCT, randomised control trial,reps; repetition, RM; repetitive movement, ROM; range of motion, sig; significant, TUG; timed up and go test, w; week

Table 2.5 Studies of aerobic training in young people with cerebral palsy

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Lunderberg et al. (1967)	Pre-post test	n=14,15–20 years, GMFCS not specified	1) Before 2) After 6 weeks' community exercise, 2x/week, physio, community gymnasium, training programme: Targeting large muscle group-Running, jumping, parallel bars	1) VO_2max (cycle ergometer-submaximal test), pulse rate and blood lactic acid	1) Increase aerobic capacity (no statistical analysis was used)
Berg (1970)	Pre-post test	n=22, 7–25 years, GMFCS not specified	1) Before 2) After 3) 3mo f/up, 1.5–6 months programme, 3x/week, physio, laboratory, training programme: Bicycle ergometer at varying intensity: 20 mins	1) muscle strength 2) VO_2max–bicycle ergometer	1) Improvement in muscle strength 2) increase in maximal heart rate (no statistical analysis were used)
Shinohara et al. (2002)	Case series	n=11,13.3–15.8 years (mean not specified), GMFCS not specified	1) Before 2) During 3) After 6–20-week programme, 2x/week, not described, laboratory, training programme: cycle ergometer or arm cranking at the anaerobic threshold point for 20 mins	1) Oxygen uptake (cycle ergometer), Interview of the children	1) Sig improvement in oxygen uptake at the anaerobic threshold point increased significantly in the leg exercise group ($p<0.05$)

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurements, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Schlough et al. (2005)	Case series	n=3, 17–20 years (mean 18.3),GMFCS (I=1), III=2)	1) Before 2) During 3) After 4) f/up, 10–21weeks exercise, 3x/week, not described, community gymnasium, training programme: Treadmill, elliptical, recumbent stepper	1) Isometric strength of quadriceps, hamstrings, ankle plantarflexor, and dorsiflexor (handheld dynamometer) 2) GMFM 3) EEI 4) self-perception profile (SPPCS)	1) No sig change 2) No sig change 3) Mixed results 4) No sig change

EEI; energy expenditure index, f/up; follow up, GMFCS; Gross Motor Function Classification System, GMFM; Gross Motor Function Measure, mins; minutes, mo; months, Physio; physiotherapy, sig; significant, VO_2max; maximal oxygen uptake

Table 2.6 Studies of strength and aerobic training in young people with Cerebral Palsy

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurement, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Darrah et al. (1999)	Pre-post test	n=23, 11–20 years (mean 14.2), GMFCS not specified	1) Before 3x 2) 10w 3) 20w f/up, 10-week community programme, 3x/week, physio student, community gym, training programme: aerobic exercise (progress from 10–13 min throughout the programme), 3 sets of 12 reps for weight training circuit of upper and lower limb	1) Isometric muscle strength (handheld dynamometer) 2) EEI& submaximal heart rate (cycle ergometer) 3) Flexibility (sit and reach test) 4) Self-perception profile	1) Sig improve post training and f/up $p<0.01$ 2) No sig change 3) No sig change 4) No sig change
Unnithan et al. (2007)	RCT	n=13,14–18 years (mean 15.8), GMFCS II=4),III=9)	1) Before 2) After,12w, 3x/week, physio, rehab centre, training programme: 5 sets of 10 reps strength training upper limbs, trunk, abs and lower limbs, aerobic interval training for 20–22mins (uphill walking reps), Both groups maintain the normal physio intervention 2x/week	1) Submaximal heart rate (VO_2max at 4 min max workload using arm cranking) 2) Aerobic capacity (VO_2max at plateau at the end of the test-spirometry) 3) GMFM	1) Sig difference between group post training $p<0.05$ 2) Sig difference between group post training $p<0.05$ 3) Sig improve in exs group $p<0.05$

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurement, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Verschuren et al. (2007)	RCT single-blind	n= 65, 7–18 years (mean 11.6),GMFCS (I=47,II=21)	1) Before 2) 4 mo 3) 8 mo 4) 12 mo f/up, 8-month exercise training, physio, community gym, training programme: 25 to 30 mins functional aerobic, anaerobic and muscle-strengthening exs, Control group-maintain the normal rehabilitation care	1) Aerobic capacity (10-m SRT) 2) Anaerobic capacity (Muscle Power Sprint Test) 3) Agility (10x5M Sprint Test) 4) Muscle strength (30 sec reps max) 5) Self-concept (SPPC) 6) GMFM 7) Participation (CAPE) 8) Quality of life (TACQOL)	1) Sig difference between group post training p<0.01 2) Sig difference between group post training p=0.04 3) Sig difference between group post training p<0.01 4) Sig difference between group post training p<0.01 5) Sig difference between group post training (athletic p=0.05) 6)Sig difference between group post training dimension D p=0.03 7)Sig different between group post training overall p=0.02 8)Sig different between group

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurement, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
					post raining (basic motor function, cognitive p<0.05)
Fowler et al. (2010)	RCT single-blind	n=62, 7–18 years (mean 11.4), GMFCS (I=19,II=14),III =29)	1) Before 2) After 12-week exercise programme, physio, 3x/week, community-paediatric physio clinic training programme: Each session of 60mins cycling inclusive of lower limbs strengthening and cardiorespiratory endurance, Control group- maintain normal physical activity	1) 600-yard walk run test 2) 30 secs walk test 3) GMFM D&E 4) Peak extensor and flexor isometric and isokinetic moments	1) Sig improvement in exs group p=0.03 2) No sig change 3) No sig change 4) Sig improvement in exs group knee extensor moment at 120^{o}/s and knee flexor moment at 30^{o}/s p<0.05
Demuth et al. (2012)	As above	As above	As above	1) PedsQoL& PODCI	1) Sig difference within group in emotional functioning (PedsQL) and treatment expectations (PODCI-parents) p<0.05

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurement, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
Slaman et al. (2014)	Multi-centre RCT	n=57 16 to 24 years (20 years) GMFCS (I=33, II=18, III=5, IV=1)	1) Before 2) 14w 3) 26w 4) 1-yr f/up, 6-months exercise programme, physio, fitness centre & home, training programme: 1) Weekly supervised centre and weekly home-based fitness training (aerobic and strength) for 3 months 2) Counselling on daily physical activity for 6 months, 30 mins duration per session 3) Counselling on sports participation, Control group: continue regular physiotherapy treatment	1) Cardiopulmonary fitness 2) muscle strength 3) body composition	1) Sig improvement in VO_2max on the anaerobic threshold after 3 months programme 2) No sig change 3) Sig improvement at 12 month follow-up in experimental group compared to control
Slaman et al. (2014)	As above	As above	As above	1) Fatigue 2) Social participation 3) Quality of life (SF36) 4) GMFM	1) Sig improvement post intervention $p<0.05$ 2) No sig change 3) Sig improvement in (mental

Study (Author and Year)	Design	Participants (n, age, GMFCS)	Programme details (Time and number of measurement, programme duration, frequency of the exercise, supervision, venue, training programme and exercises, other details)	Outcome measures used	Result for each outcome measure
					health) and family support (participation and involvement) post intervention and bodily pain, mental health) and family support (participation and involvement) at 1-yr f/up
Slaman et al. (2015)	As above	As above	As above	1) Objective measurement of movement behaviour (VitaMove system) 2) PASIPD	1) No sig change 2) No sig change

CAPE; Children Assessment of Participation and Enjoyment, EEI; energy expenditure index, exs; exercise, f/up; follow up, GMFCS; Gross Motor Function Classification System, GMFM; Gross Motor Function Measure, mins; minutes, mo; months, PASIPD, Self-reported physical activity (The Physical Activity Scale for Individuals with Physical Disabilities, PedsQoL; Pediatric Quality of Life inventory, PODCI; Pediatric Outcomes Data Collection Instrument, reps; repetition, sig; significant, SPPC; self-perceived communication competence scale, TACQOL; TNO-AZL (Netherlands Organisation for Applied Scientific Research Academic Medical Centre) Children's Quality of Life questionnaire, VO_2max; maximum oxygen uptake, w; week, yr; year

2.3.3 Gaps in the evidence and rationale for the studies included in this thesis

There is a growing body of evidence with regards to exercise programmes for people with CP, and this has been discussed in relation to strength exercise, aerobic exercise and combined exercise programmes in section 2.3. The literature diverges in methodologies and results and this has been summarised and presented in Tables 2.4 to 2.6. The majority of the current evidence reveals that exercise programmes for people with CP have positive outcomes on one or more aspects of physical function. However, further investigations would add to our knowledge and should attempt to address some of the following gaps in the literature:

- The majority of the studies (15 out of 25 studies) included children with CP (mean age < 16), with little attention paid to young people aged between 16 and 25 years. Future studies should thus include those who are just prior to entering and following transition to adult physiotherapy services.
- A follow-up assessment following completion of the exercise programme is important for assessment of whether any benefits of the intervention have been sustained as well as to establish knowledge on participants' physical activity or participation in exercise; therefore, this should be included in future studies.
- Utilising valid and reliable outcome measures to assess the impact of an intervention on short- and long-term physical activity levels.
- As a majority of the studies undertaken are small in size (only 7 out of 25 studies with sample size n>30) and therefore often underpowered, larger, appropriately powered studies should be undertaken.
- Implementing the exercise programme at a community leisure centre may allow increased participation and should be included in future work.
- Only one study (McBurney et al. 2003) used in-depth interviews exploring the participants' thoughts regarding the exercise

programme, which may provide fruitful feedback regarding the programme (i.e. why such a programme was/was not a success or sustainable); thus, qualitative methodologies should be included to establish this.

2.3.4 Summary

There is scope for continued clinically relevant research in exercise for young people with CP using high-quality study design, and this will contribute to the evidence for effective care and treatment options for those with CP. So far, however, there is limited evidence with regard to the effects of a pragmatic community exercise programme for young people with CP beyond the age of 16 in the UK. The majority of the existing research studies paid more attention to children with CP who were below 16 years of age. Therefore, the intervention study (Study 3, Chapter 7) sought to address the gaps in the literature, that is, by conducting an exercise programme targeting individuals with CP between 16 and 25 years of age, a follow-up assessment following completion of the programme, using a large sample size and implementing the programme at a community leisure centre.

The next section (2.4) will provide a literature background to the OMs used in the test-retest Study 2 (Chapter 5) and intervention Study 3 (Chapter 7).

2.4 Outcome measures to be used in exercise intervention studies involving young people with cerebral palsy

There are many OMs used in clinical and research settings when assessing people with CP as well as evaluating the effects of exercise intervention in this population. This section will explore the basic concept of the psychometric properties of OMs followed by the relevance of the OMs with regard to the International Classification and Functioning (ICF) (World Health Organization 2007). Then, the description, validity and reliability, feasibility and future work of the OMs chosen for this thesis will be discussed in detail.

2.4.1 Psychometric properties of outcome measures – A basic concept

To objectively measure the impact of an exercise intervention, quantitative measurement is commonly used. There are a number of ways to achieve this; for example, performance-based tests, instrumental tools or self-reported questionnaires (Patient Reported Outcome measures or PROMS). Apart from being valid, reliable and responsive to change, OMs must demonstrate their practicality (feasibility) when used in clinical or research settings where appropriate. The elements that contribute to the statistical adequacy of an instrument in terms of its reliability, validity, measurement error, responsiveness and internal consistency are known as its psychometric properties. Clinimetrics is a measurement of patients through clinical data involving scales, indexes and quantitative instruments (Gabel et al. 2012).

2.4.1.1 Reliability

According to Mokkink et al. (2013), reliability is the extent to which scores for patients who have not changed are the same across repeated measurements. Within the reliability domain there are several psychometric properties, which are: test-retest, inter-rater reliability, intra-rater reliability, measurement error and internal consistency. When the consistency of an OM is compared over time it is known as the test-retest reliability. Inter-rater reliability is established when comparing the outcome by different raters on the same occasion. Intra-rater reliability is assessed when comparing the results from the same rater on different occasions. Intraclass correlation (ICC) and weighted Cohen's Kappa are the most common and appropriate statistical parameters used for continuous and ordinal scores, respectively, to assess 'consistency' when investigating the test-retest, inter-rater reliability and intra-rater reliability of a particular OM (Terwee et al. 2007). An ICC value >0.75 indicates good reliability, 0.50–0.75 indicates moderate test-retest reliability and <0.50 represents poor test-retest reliability (Portney 2000). According to Streiner and Norman (2008), Pearson correlation is not suitable for use as it measures the linear relationship and not systematic

differences. Measurement error, a measure of ‘agreement’, is the systematic and random error of a patient’s score that is not attributed to true changes in the construct to be measured (Mokkink et al. 2013). Examples of indices of measurement error are standard error of measurement (SEM) (Stratford 1989), minimum detectable change (MDC) and limit of agreement (Bland and Altman 2007). Internal consistency is the degree of the interrelatedness among the items of an instrument, which is mostly relevant for questionnaires. Cronbach’s alpha is an adequate parameter to express internal consistency (Terwee et al. 2007).

2.4.1.2 Validity

Apart from reliability, another important psychometric property of an OM that should be established is its validity. Validity can be divided into content, construct and criterion (Mokkink et al. 2013). Content validity is relevant to questionnaires as it examines the extent to which the concepts of interest are comprehensively represented by the items in the questionnaire (Guyatt 1993). There is no statistical parameter with which to measure content validity; however, according to Terwee et al. (2007), questionnaires should provide the measurement aim, target population, the concepts that are being measured and the item selection. In addition, the target population should have been involved during item selection, as well as either investigators or experts to establish content validity. The degree to which the scores of OMs are consistent with the hypotheses (for instance, with regard to internal relationships, relationships to scores of other instruments or the differences between relevant groups) is known as construct validity (Mokkink et al. 2012). The Pearson correlation coefficient (r) is the parameter used to represent correlation. The strength of the correlation is regarded as small if r has a value in the range 0.1– 0.3, moderate if r falls within 0.3–0.5 and strong if r is within the range 0.5–1.0 (Portney and Watkins 2000). Criterion validity refers to the extent to which the scores on a particular instrument relate to a gold standard (Terwee et al. 2007).

2.4.1.3 Responsiveness

Responsiveness is defined as the ability of an OM such as a questionnaire to detect clinically important changes over time, even if these changes are small (Guyatt et al. 1989). Terwee et al. (2003) suggested that responsiveness is a measure of longitudinal validity. Analogous to construct validity, longitudinal validity should be assessed by testing predefined hypotheses, e.g. about expected correlations between changes in measures, or expected differences in changes between 'known' groups (Terwee et al. 2003). This shows the ability of an OM to whether changes really taken place. Furthermore, the OM should be able to distinguish clinically important change from measurement error. There are various statistical approaches to calculating responsiveness (Crosby et al. 2003). According to Mokkink et al. (2013), the standardised response mean and effect size of the OM under investigation are inadequate to calculate responsiveness as they measure the effectiveness of the intervention rather than the quality of these measurement properties. Instead, these authors recommended analysing the correlation coefficients between the OM under investigation against another established OM (often the gold standard and ideally a patient-reported questionnaire) or by calculating the Receiver Operating Curve (ROC) to calculate the sensitivity or specificity of the OM in question.

The parameter used to express responsiveness is the Minimum Clinically Important Difference (MCID). Any amount of change greater than the MCID threshold is considered to be clinically meaningful or important. The MCID is a concept that is more clinically oriented and is focused at the level of the individual patient. These concepts are important given that statistically significant change at the group level may not be clinically significant at the individual level, or vice versa. Further, average effects across a group may not be meaningful to interpret the degree of change in an individual patient (Schmitt and Di Fabio 2004, Guyatt et al. 2002). The anchor-based approach is the method recommended to quantify MCID (de vet et al. 2006). In the anchor-based approach, the magnitude of change in the OM under

investigation is compared with a more established OM or patient-perceived change in health status following the treatment. In the distribution-based approach, the variability of the OM itself is calculated, and this could be done by calculating the SEM or effect size of the score distribution (Jaeschke et al. 1989).

2.4.2 Outcome measure – linked with International Classification of Functioning

The outcome measures (OMs) chosen in the current study can be categorised using the International Classification of Functioning (ICF) (World Health Organization 2007). The ICF is a model for describing health-related conditions and their impact on the individual. Moreover, it provides a universal framework for defining and classifying functioning and disability with its domains; body function and structures, activities and participation, as well as contextual factors (World Health Organization 2007). Comprehensive core sets of categories based on the ICF domains have been established for CP in patients aged 0 to 18 years. The ICF core sets for CP highlights the 'what' to measure and could be used to guide the clinician and researcher when assessing their CP client. The selection of the core set to be used will vary depending upon the intended purpose and setting. This means that the choices of OMs to be used can be variable when measuring the outcome in people with CP. It was also suggested that the selection of appropriate OMs depends on several factors, as follows: 1) the research questions, 2) the type of intervention, 3) the content and psychometric properties of the OMs, and 4) the characteristics of the target population, for example, age (Schiariti et al. 2014). Hence, the current study has considered those factors when choosing the OMs to be used in the intervention study (Chapter 7). The OMs used in the exercise intervention study in relation to ICF are shown in Table 2.7. The details of each OM inclusive of psychometric properties, rationale and content will be explained in the next section.

Table 2.7 Outcome measures used in this study

ICF domains	Code	ICF category name	Outcome measures used in this study
Body functions	b455	Exercise tolerance functions	Shuttle Run test
	b710	Mobility of joint functions	Photographic passive range of motion
	b730	Muscle power functions	Isometric muscle strength
	b770	Gait pattern functions	Three-Dimensional Gait Analysis
Activity and participation	d4	Mobility	Gross Motor Function Measure Gillette Functional Assessment Questionnaire Canadian Occupational Performance Measure Objective Habitual Physical Activity (ActivPAL^{tm})
	d450	Walking	Timed Up Go test
	d9	Community, social and civic life	Canadian Occupational Participation Measure Short Form 12
Personal factors	-	-	Rosenberg Self-Esteem Scale

2.4.3 Outcome measures used in this study

Previous studies have used many different OMs to measure the effects of an exercise programme, as shown in Table 2.8. These vary in terms of the construct measured, their psychometric properties and their feasibility of use in either clinical practice or a research setting. A discussion on the different OMs used in Study 3 (Chapter 7) and, where relevant, other similar OMs in past studies, will follow. The reader is also referred to section 2.3 and Tables 2.4–2.6, where the previous exercise studies and the OMs used can also be

found. If studies evaluated the same outcome measure and were sufficiently homogenous (i.e. study population, design, measurement procedure, rating), their overall evidence was rated on the basis of the Cochrane Back Review Group Criteria as ‘strong’, ‘moderate’, ‘limited’, ‘conflicting’ or ‘unknown’ (van Tulder et al. 1976). The details of the Cochrane Back Review Group Criteria are as follows:

a. strong – consistent findings in multiple studies of good methodological quality OR in one study of excellent methodological quality

b. moderate – consistent findings in multiple studies of fair methodological quality OR in one study of good methodological quality

c. limited – one study of fair methodological quality

d. conflicting – conflicting findings

e. unknown – only studies of poor methodologies.

Table 2.8 List of outcome measures used in adolescents and young adults with cerebral palsy in previous exercise studies

ICF domain	Parameter/ Construct	Outcome measures	References of studies in which OM is used
Body functions	Aerobic capacity	Oxygen uptake	Lunderberg et al. (1967), Berg (1970), Shinohara et al. (2002),
		10-m shuttle run test	Verschuren et al. (2007)
		VO_2 max (maximum oxygen uptake)	Unnithan et al. (2007), Slaman et al. (2014a)
	Anaerobic capacity	Muscle power sprint test	Verschuren et al. (2007)
	Muscle strength	Dynamometry (manual or instrumented)	MacPhail and Kramer (1995), Dodd et al. (2003), Eagleton et al. (2004), Taylor et al. (2013), Schlough et al. (2005), Darrah et al. (1995), Slaman et al. (2014a)
	Flexibility	Sit and reach test	Darrah et al. (1995)
	Range of motion	Goniometer	Healy (1958)
	Muscle tone	Modified Ashworth Scale	MacPhail and Kramer (1995), Patikas et al. (2006)
	Gait kinematics	Gait Profile Score	Unger et al. (2006), Patikas et al. (2006) Taylor et al. (2013),
Activity and participation	Gross motor function	Gross Motor Function Measure	MacPhail and Kramer (1995), Patikas et al. (2006), Taylor et al. (2013), Schlough et al. (2005), Unnithan et al. (2007), Verschuren et al. (2007), Slaman et al. (2014b)
	Cadence	Visual observation	Eagleton et al. (2004)
	Walking efficiency	Energy expenditure index	MacPhail and Kramer (1995), Eagleton et al. (2004), Schlough et al. (2005), Darrah et al. (1995), Patikas et al. (2006),
	Walking speed over short distance	Self-selected walking speed	Dodd et al. (2003)
	Walking speed in endurance test	6-minute walk test	Taylor et al. (2013)
	Functional capacity in Activities daily living	Time stair test	Dodd et al. (2003)

ICF domain	Parameter/ Construct	Outcome measures	References of studies in which OM is used
	Functional mobility	Functional Mobility Scale	Taylor et al. (2013)
		Functional Assessment Questionnaire	Taylor et al. (2013)
	Physical activity and sedentary time	Body-fixed accelerometer	Slaman et al. (2015)
	Self-reported physical activity	The physical activity Scale for Individuals with Physical Disabilities	Slaman et al. (2015)
	Participation	The CAPE	Verschuren et al. (2007)
		Life-H	Slaman et al. (2014b)
	Quality of life	TACQOL	Verschuren et al. (2007)
		PedsQOL	Engsberg et al. (2006)
		SF36	Slaman et al. (2014b)
	Fatigue	Fatigue Subscale Fatigue Severity Scale	Slaman et al. (2014b)
Personal factors	Self-perception	Self-perception Questionnaire	Unger et al. (2006)
		Self-perception profile	Schlough et al. (2005), Darrah et al. (1995), Verschuren et al. (2007), Dodd et al. (2004)
		Perception on body image, functional performance, social participation (semi-structured interview)	McBurney et al. (2003)

CAPE; Children Assessment of Participation and Enjoyment, Life H; Assessment of life habits, PedsQoL; Pediatric Quality of life inventory, SF36; Short form 36, TACQOL;TNO-AZL (Netherlands Organisation for Applied Scientific Research Academic Medical Centre) Children's Quality of Life questionnaire

2.4.3.1 Isometric muscle strength

Muscle weakness is common in CP (Malaiya et al. 2007, Lampe et al. 2006, Thompson et al. 2011). Decreased muscle strength in children with CP is often complicated by other factors such as spasticity (Ross and Engsberg 2007a) and can lead to joint contractures that cause pain and fatigue (Vogtle et al 2013), limitations in walking (Desloovere et al. 2006, Ross and Engsberg 2007b) and reduced functional activity (Damiano and Abel 1998).

Previous CP studies mostly used a handheld dynamometer to detect changes in muscle strength post exercise training (McPhail and Kramer 1995, Dodd et al. 2003, Eagleton et al. 2004, Taylor et al. 2013, Schlough 2005, Darrah et al. 1995). However, the measurements obtained when using a handheld dynamometer are dependent on the strength of the assessor. Alternatively, a fixed digital dynamometer allows the assessor to securely attach one end to a fixation point, thus removing the need for the assessor to

resist the muscle force. The fixed digital dynamometer has been used previously in children and adolescents with CP and has been found to be able to measure difference in muscle strength between ambulant CP children and their healthy peers (Thompson et al. 2011). An alternative method for measuring muscle strength lies in the use of sophisticated isokinetic testing equipment; however, this is expensive and is often not available in clinical and research settings (Berg-Emons et al. 1996).

Validity and reliability of handheld and fixed digital dynamometer

The reliability of the handheld dynamometer for measuring isometric strength has been reported in a study of participants with brain injury (Riddle et al. 1989). In particular, Riddle et al. (1989) reported good consistency, with an ICC of 0.9–0.96 (95% CI was not reported). It was also found to be reliable in healthy and CP children with ICCs of 0.84 (95% CI was not reported) (knee extensors) and 0.79 (hip extensor) (Seniorou et al. 2002). The test-retest reliability of the measurement of gluteus maximus strength using a fixed digital dynamometer was found to be good in a group of CP patients aged 6–14 years with ICCs ranging from 0.75 to 0.83 (95% CI was not reported) (van der Linden et al. 2004). The correlation between a fixed handheld dynamometer and isokinetic dynamometer for isometric knee extensor strength was found to be strong ($r=0.806$, $p<0.05$) in a normal adult population (Kim et al. 2014). The study also found that the inter-rater and intra-rater reliabilities of the fixed handheld dynamometer were high, with ICCs ranging from 0.95 to 0.98 for knee extensor strength (Kim et al. 2014).

Feasibility

Standardised protocols are important when assessing the efficacy of treatment. Therefore, for Study 2 (Chapter 5) and Study 3 (Chapter 7), the protocol outlined by Thompson et al. (2011), the myometer (MIE Medical Research Ltd), was used.

Future work

There is moderate evidence regarding the reliability of the fixed digital dynamometer in children with CP; however, this is not the case for adolescents. Therefore, isometric muscle testing using a fixed digital dynamometer was chosen for the test-retest reliability study (Study 2, Chapter 5) and the exercise intervention in Study 3 (Chapter 7), in order to address the gap in the present evidence.

2.4.3.2 10m Shuttle Run Test (SRT)

Aerobic capacity is defined as the ability to deliver oxygen to the muscles and to utilise it to produce energy during exercise (Armstrong and Welsman 2007). High levels of aerobic capacity are shown to prevent health problems as a result of inactivity (Armstrong and Welsman 2007, Carnethon et al. 2005), increase participation (Majnemer et al. 2008) and facilitate the completion of activities of daily living (Bjornson et al. 2008).

Verschuren et al. (2007) used an adapted 10m shuttle walk/run test (10m SRT) to measure aerobic capacity in their study, including that of children and adolescents with CP. The usage of the 10m SRT is a more economical and practical method of assessing maximum aerobic capacity compared to the use of a treadmill in laboratory tests.

The SRT was originally developed by Leger et al. (1988) to measure aerobic fitness in healthy adults. Verschuren et al. (2006) made some modifications to the SRT, specifically to measure aerobic fitness in children and adolescents with CP GMFCS levels I and II and, later, level III (Verschuren et al. 2011). This modification for CP requires participants to walk/run/jog between two markers that are 10m apart, compared to the original SRT in which the markers are 20m apart. The participants all walk/run/jog at the same time, using a pre-recorded bleep sound played from a standard audio compact disc (CD) player. The audio CD for those with GMFCS I and II is

different in terms of the starting speed; however, it has the same increment rate of 0.25km/hour approximately every minute. Participants with GMFCS level I start at 5km/hour while those with GMFCS level II begin at 2km/hour. Participants with GMFCS level III using the same audio CD as those with GMFCS II; however, these participants are required to walk/run/jog along the sides of a 7.5-metre square (thus requiring only a 90^{o} rather than a 180^{o} turn at each bleep) (Verschuren et al. 2011). At the end of each level, the participants are instructed to go a bit faster. The test ends if the participants are unable to keep going or fail to reach the next marker before the next bleep twice in a row.

Validity and reliability

The validity and reliability of the 10 m SRT in participants with CP aged 7–17 GMFCS I and II have been reported by Verschuren et al. (2006). The authors reported that the test-retest reliability of peak heart rate derived from the 10m SRT was good, with ICCs of 0.97 and 0.94 (95% CI was not reported) for those with GMFCS I and II, respectively. The test-retest reliability in GMFCS III was also found to be good, with an ICC of 0.98. In terms of validity, peak oxygen uptake (VO_2max) obtained from the 10m SRT and from a graded treadmill showed a strong correlation ($r=0.96$, $p<0.01$) for children with both GMFCS I and II (Verschuren et al. 2006).

Feasibility

The 10m SRT is a practical and preferable method to the measure estimation of maximal oxygen uptake using a treadmill (Verschuren et al. 2006). It requires space for a greater than 10m run/walkway and also for the participants to make a turn for the next walk or run. The only other equipment required are a CD and CD player (Verschuren et al. 2006) and two or four markers (i.e. cones) to mark the distances for GMFCS I/II or GMFCS III, respectively.

Further work

There is limited evidence regarding the reliability of the 10m SRT in young people aged >16 years with CP as so far only one reliability study has been reported in children with CP aged 7–17. Thus, the test-retest reliability of the 10m SRT was assessed as part of Study 2 (Chapter 5) and the exercise intervention in Study 3 (Chapter 7).

2.4.3.3 Timed Up and Go Test

The Timed Up and Go test (TUG) was originally designed to measure functional mobility in the elderly population (Podsiadlo and Richardson 1991). The TUG is a very simple and easy test that requires participants to stand up from a chair and walk at a normal pace around a cone that is placed 3 metres from the chair, prior to returning and sitting back on the same chair. The time taken to complete the test is recorded using a standardised digital timer.

Validity and reliability

Williams et al. (2005) found good consistency of the TUG, with an ICC of 0.99 (95% CI 0.91–0.99), in a study of 33 participants with CP aged 3 to 19 years. The study also found moderate negative correlation between TUG and GMFM (rho=-0.524, p=0.012). In addition, they found that TUG scores differed between young people with CP classified at GMFCS I, II and III. Similarly, Dhote Sanjivani (2012) reported a good ICC (0.99) (95% CI was not reported) in test-retest reliability in a study of children with CP aged 4–12 years.

Feasibility

A very easy, simple and quick test to measure functional mobility with minimal equipment and space is needed. Equipment required: a chair with arm support, timer and cone (to mark the 3-metre distance).

Further work

There is moderate evidence as to the psychometric properties of TUG in populations with CP; hence, further work on the reliability and validity of the TUG across the age range in CP is required. To confirm the results of previous work on the reliability of the TUG in adolescents with CP, the TUG was included in Study 2 (Chapter 5). The TUG is also utilised in the exercise intervention in Study 3 (Chapter 7).

2.4.3.4 Three-Dimensional Gait Analysis

Gait is the pattern movement of the limbs in humans during locomotion. Gait analysis provides a useful understanding of basic walking ability and abnormalities in pathological gait. There are various methods of gait analysis, comprising visual observation, video recording and computerised 3DGA (Levine et al. 2012). Visual gait analysis has several disadvantages as there is no permanent record; it thus depends on the skills of the observer and is subjective, which sometimes gives rise to bias if the observer is non-blind to the patient's history. However, it should be acknowledged that visual gait analysis is beneficial in a clinical setting as there is no additional cost, space or time involved. Video-based observation allows the clinician to record the patient's walking and thus overcome the issue of the lack of a permanent record and the difficulty of observing high-speed events. Several OMs can be used to score video-based observation, including the Edinburgh Visual Gait Score, Physician Rating Scale and Visual Gait Assessment Score, and produce outcomes in the coronal and sagittal planes during the swing and stance phase. In order to understand more complex gait pathologies or gain insight into the underlying mechanisms of abnormal patterns, a more complete motion analysis that measures kinematics and kinetics can be useful, and this can be achieved through the use of the 3DGA. The 3DGA method provides temporal-spatial parameters such as cadence, step length and walking velocity, in addition to kinematic (i.e. joint angles) and kinetic parameters (i.e. joint forces and moments) in all three planes of movement. Possibly the most common 3DGA system is Vicon (Oxford Metrics, Oxford,

United Kingdom). Other motion analysis systems that have been used in other CP studies are OrthoTrak (MotionAnalysis Corporation, Santa Rosa, CA, USA) and Optotrak (Northern Digital, Waterloo, Canada) (Mackey et al. 2005, Grunt et al. 2010). For the current study, the Vicon 3DGA system was used, as the laboratory at Queen Margaret University is equipped with this system.

Validity and reliability

The reliability of spatial-temporal gait parameters and joint kinematics derived from 3DGA has been explored in many studies involving participants with CP (Mackey 2005, Redekop 2008, Klejman 2010, Steinwender 2000). Redekop et al. (2008) investigated test-retest reliability and reported good ICCs of between 0.76 and 0.84 (95% CI 0.57–0.95) for spatial-temporal gait parameters in children with CP with GMFCS I and a mean age of 8 years, one month. The test-retest results in a study of participants with CP aged 5 to 16 showed ICCs for joint kinematics in the sagittal plane to be between 0.93 and 0.99 (95% CI was not reported) (Mackey et al. 2005). A study of 17 young people with CP aged 4 to 14 revealed that the intra-rater reliability of sagittal kinematics ranged between moderate and good (ICCs 0.58–0.99) (95% CI was not reported) and inter-rater reliability was between poor and good (ICCs 0.48–0.99; 95% CI was not reported) (Grunt et al. 2011). The means and standard deviations of coefficient of variation percentage for the cadence, stride length within days, were reported to be 3.9 (2.8)%, 6.6 (4.6)% and 5.0 (3.4)% respectively in a study of 20 children with spastic diplegic between the ages of 7 and 15 years (Steinwender et al. 2000). A full appraisal of the psychometric properties of the 3DGA used in young people with CP are discussed in Study 1 (Chapter 4).

Feasibility

Measurement of joint kinematics, joint kinetics and spatial-temporal parameters using 3DGA systems produces quantitative data on gait patterns which other walking tests such as visual and video-based observations

cannot produce. However, the set-up (marker placement especially) is time-consuming and training/familiarisation with the equipment is needed. Since QMU has a Vicon motion-analysis system, 3DGA was used for gait analysis in Study 3 (Chapter 7).

Future work

Clearly, the literature showed that there is strong evidence regarding the reliability of 3DGA. To further strengthen the current evidence, joint kinematics, as derived from 3DGA, will be included in Study 3 (Chapter 5).

2.4.3.5 Gross Motor Function Measure 66

Dimensions D and E in the GMFM represent areas that many young people with CP who are able to walk have difficulty with (Damiano et al. 1995). Unsurprisingly, most of the previous exercise intervention studies chose these dimensions when assessing their participants' gross motor function (MacPhail and Kramer 1995, Patikas et al. 2006, Taylor et al. 2013, Schlough 2005, Unnithan et al. 2007, Verschuren et al. 2007). Well-established protocols for administering the GMFM, as well as training material on how to score the measure, can be found in the user's manual (Russell et al. 2002). Scores are presented as percentages. Separate scores can be calculated for each of the five dimensions as well as for the total scores (Russell et al. 2002). Although the GMFM has been validated for use with patients with CP aged 4–18 years, it has also been used in an exercise study of CP with participants aged 25–47 (Andersson et al. 2003).

The GMFM has been widely used in CP exercise intervention studies (MacPhail and Kramer 1995, Patikas et al. 2006, Taylor et al. 2013, Schlough 2005, Unnithan et al. 2007, Verschuren et al. 2007). It is used to measure the change in gross motor function in children with CP (Russell et al. 2002). GMFM has five dimensions, as follows: (A) lying and rolling, (B) sitting, (C) crawling & kneeling, (D) standing, and (E) walking, running and jumping. Items are scored using a four-point Likert scale (0, could not initiate task; 1,

initiated task (<10% of task); 2, partially completed task (10 to <100%); 3, completed task). GMFM 66 is the most recent version and comprises 66 items and is only suitable for use with children with CP (Russell et al. 2002). The original version, known as the GMFM 88, contains 88 items and has been validated for children with CP and Down's syndrome (Russell et al. 2009). It can also be used in children with osteogenesis imperfecta and acute lymphatic leukaemia (Russell et al. 2009). The differences between the GMFM 66 and 88 were described in a study of a group of children with CP who underwent a selective dorsal rhizotomy operation (Josenby et al. 2009). This study revealed that the total scores for the GMFM 88 indicated large changes in motor function sooner after the surgery in comparison with the GMFM 66 scores in children with GMFCS levels I–III and IV–V (Josenby et al. 2009).

Validity and reliability

The GMFM has been shown to be valid, reliable and responsive through a number of studies looking at motor status in CP, and to be able to quantify changes over time or as a result of intervention (Vos-Vromans et al. 2005, Russell et al. 2010, Nordmark et al. 2000, Oeffinger 2008). Bjornson et al. (2000), in a study of 21 participants with CP aged 4 to 18 years, reported that the ICC of the test-retest was good (0.99) (95% CI was not reported) for dimensions D and E. In a different study, the inter-rater reliability (ICC) was also found to be 0.99 (good) (95% CI 0.99) and the correlation between GMFM and video-based evaluation was strong ($r=0.82$, $p<0.05$) (Russell et al. 2012). Moderate correlation ($r=0.42$ and 0.43, $p<0.05$) between GMFM dimensions D and E and global rating scale for parents was reported in children with CP greater than 4 years of age. In addition, the effect size was moderate (0.47 and 0.67) in dimensions D and E respectively (Vos-Vromans et al. 2005).

Feasibility

The GMFM 66 is very easy to administer, although it can be time-consuming. It can take approximately 45 to 60 minutes for someone familiar with the measure to complete, depending on the skill of the assessor, the ability level of the participant and the participant's level of cooperation and understanding. The GMFM user manual provides very clear instructions on how to use it. As our participants were ambulant CP patients, we tested them only for dimensions D (standing) and E (walking, running and jumping) (Appendix 1).

Future work

The previous literature has shown there is strong evidence of the GMFM's reliability; however, only one study included adolescents, with the majority of the studies focusing on children. Therefore, we included the GMFM 66 when measuring the outcome of exercise intervention in Study 3 (Chapter 7) to add to the currently available knowledge.

2.4.3.6 Objective habitual Physical Activity

Activity monitors are recommended since they provide more objective information about participants' habitual daily physical activity. Participants can wear an activity monitor during their activities of daily living. Clanchy et al. (2011), in their review of objective measures for physical activity in adolescents with CP, reported HR flex, StepWatch, accelerometers and pedometers as the most commonly used devices among this population. Another activity monitor used in this population is the ActivPAL^{TM} (PAL Technologies Ltd, Glasgow, Scotland) (Grant et al. 2006). It is a single-unit monitor based on a uni-axial accelerometer. It records episodes of walking, standing and sitting/lying, thereby allowing the measurement of both activity and inactivity. In addition, the monitor records step count, instantaneous cadence and sit-to-stand transitions.

Validity and reliability

A study in Scotland showed that ActivPAL™ demonstrated excellent ability to distinguish between the sitting/lying and upright postures in a CP population (Tang et al. 2013). The Scottish study involved 15 ambulant children and adolescents with CP by comparing the ActivPAL™ with video analysis. The results showed that the agreement was (mean±SD) between 97.4 ± 2.7% and 103.8 ± 10.1% (Tang et al. 2013). McAloon et al. (2014) compared the time spent standing and walking, number of steps taken and the number of transitions derived from the ActivPAL™ with observations from video analysis. They found a high agreement between the two methods; the absolute difference between the two methods for walking time was 2.2s and for step counts was 3.2 steps in participants with CP hemiplegia aged between 4 and 18 years. ActivPAL™ was also found to be valid in other populations, including healthy adults (Godfrey and Lyons 2007, Godfrey et al. 2007, Ryan et al. 2006, Lyden et al. 2012, Hart et al. 2011, Harrington et al. 2011) and other patient groups (Clarke-Moloney et al. 2007, Dahele et al. 2007, Egerton and Brauer 2009).

Feasibility

Users of ActivPAL™ require it to be attached to the front of the mid-thigh. The activity monitor is small in size (35 mm x 53 mm x 7 mm) and light in weight (20 mg). It is capable of recording time spent sitting/lying, standing and walking for a maximum of 10 days.

Future work

The previous literature shows there is limited evidence on the reliability of ActivPAL™ in the CP population. Hence, ActivPAL™ was included for the test-retest in Study 2 (Chapter 5) and the exercise intervention in Study 3 (Chapter 7).

2.4.3.7 Gillette Functional Assessment Questionnaire (FAQ)

Considering the importance of information obtained from participants and self-evaluation in terms of functional mobility after an exercise programme, the FAQ was used. The FAQ is a self-reported questionnaire with a ten-level classification of walking and 22 functional mobility activities on a five-level Likert-type difficulty scale (Appendix 2).

Validity and reliability

The inter-rater and intra-rater reliabilities of FAQ were found to be excellent in a CP study with good ICC (0.94, 95% CI 0.89–0.97) (Gunel et al. 2010). The correlation between FAQ and GMFCS was reported to be strong ($r=0.82$, $p<0.01$) (Gunel et al. 2010) and it was strong for the Functional Measure for Children (WeeFim) ($r=0.64$, $p<0.01$) (Novacheck et al. 2000).

Feasibility

The FAQ is a very simple questionnaire that includes questions on mobility level (one item to be selected from 10 items) and higher-level mobility skills that require participants to select things (it can be more than one) that they are able to do from a set of 15 skills (Appendix 2).

Future work

Although FAQ is widely used in the CP population, the reliability of FAQ among adolescents and young adults with CP is limited. Therefore, a test-retest reliability study of FAQ was undertaken to further strengthen the previous findings. As such, FAQ will be included in Study 2 (Chapter 5) and Study 3 (Chapter 7).

2.4.3.8 Canadian Occupational Participation Measure (COPM)

The COPM is a client-centred OM (Dedding et al. 2004) as it requires participants to identify and prioritise problems that they encounter within their daily living activities and then rate their ‘performance’ and ‘satisfaction’ with regard to those identified activities. It is a 10-point scale with a ‘performance’

rating that ranges from ‘not able to do it at all’ to ‘able to do it extremely well’ and a ‘satisfaction’ rating from ‘not satisfied at all’ to ‘extremely satisfied’ (Appendix 3).

Validity and reliability

The test-retest reliability of COPM has been found to be strong in two different studies looking at chronic obstructive pulmonary disease ($r=0.76$, $p<0.01$) and stroke patients ($r=0.89$, $p<0.01$) (Sewell and Singh 2001, Cup et al. 2003). The sensitivity of COPM was reported to be medium, with effect sizes of 0.78 and 0.69 for performance and satisfaction, respectively, in a group of 41 children with spastic hemiplegic CP three months post the occupational therapy intervention programme (Cusick et al. 2006).

Feasibility

It is easy to administer and provide flexible, consistent, individualised measures that accommodate the diverse aims of participants and exercise intervention. It may be time-consuming, especially for a first-time administrator (usually for the reassessment); however, over time it will become quicker as the participants become familiar with it.

Future work

Clearly, the reliability and validity of COPM in the CP population are unknown and limited, respectively. More high-quality psychometric properties are needed and, therefore, the COPM was included in the exercise intervention in Study 3 (Chapter 7) to fill in this gap in the knowledge.

2.4.3.9 Short Form 12 version 2 (SF-12 v2)

SF-12 v2 was used to assess the quality of life and health status of the participants in this study. This 12-item survey is a shortened version of the SF-36 (Ware et al. 1996). SF-12 v2 is a measure of general health status and measures eight domains of functioning and well-being: physical functioning, role limitations due to physical problems, bodily pain, general health

perceptions, energy and vitality, social functioning, role limitations due to emotional problems, and mental health (Ware et al. 1996). The physical and mental component scores (PCS and MCS) can be derived from the SF-12. The SF-36 has been used in previous studies that have included adults with CP (Jahnsen et al. 2004b, Opheim et al. 2009).

Validity and reliability

Cheak-Zamora et al. (2009) conducted a reliability and validity study of the SF-12 in the general population. The internal consistency of SF-12 was high (Cronbach's alpha 0.80) and the test-retest reliability for the PCS was also high (ICC=0.78) and moderate for the MCS (ICC=0.60) (95% CI was not reported). The correlation between the EuroQoL-5 dimension and PCS was strong ($r=0.56$, $p<0.01$) and moderate ($r=0.38$, $p<0.01$) with MCS (Cheak-Zamora et al. 2009). The SF-12 has been used in different CP studies for participants with ages ranging from 18 to 72 years (Jahnsen et al. 2004b, Vogtle et al. 2014, Schwartz et al. 1999).

Feasibility

It is very easy to administer with only 12 questions and requires minimal time for the participants to fill in the questionnaire (Appendix 4).

Future work

Although it has been implemented in many studies, the reliability of the SF-12 is unknown in CP populations. Therefore, the test-retest reliability of the SF-12 in young people with CP will be explored; hence, it was included in Study 2 (Chapter 5) and Study 3 (Chapter 7).

2.4.3.10 Rosenberg Self-Esteem Scale (RSES)

According to the ICF, all the domains in this model have dynamic interactions, including personal factors and those that can influence the disability and health of a person (World Health Organization 2008). Personal factors can relate to age, gender, self-esteem and coping style. Self-esteem

is defined as how one feels about oneself (Mayberry 1990). It is believed to be associated with effective functioning (Gurney 1988) and personal satisfaction (Coopersmith 1967), and understanding this aspect is crucial in planning and exploring the effectiveness of interventions for people with CP. Exercise has been reported to improve self-esteem and self-confidence in both non-disabled children and adolescents (Ekeland et al. 2005) and young people with CP (Unnithan et al. 2006). The RSES is a scale developed to measure self-esteem and was originally tested on 5,024 high school students from 10 different schools in the US (Crandal 1973, Rosenberg 1965).

Validity and reliability

The internal consistency (Cronbach's alpha) of the RSES was reported to be 0.84 in a study of 50 children aged 9 to 18 years with CP (Manuel et al. 2003). The RSES has also been found to be valid among adolescents and young adults within the healthy population (Scheier et al. 1994). An RCT conducted with a group of elderly participants (mean age of 69.6) showed that the RSES of the intervention group improved following a 16-week group exercise programme (Sung 2009).

Feasibility

The RSES is a simple questionnaire comprising 10 questions asking about general feelings and scored on a four-point Likert scale containing 'strongly agree', 'agree', 'disagree' and 'strongly disagree' (Appendix 5).

Future work

There is unknown evidence regarding the reliability of the RSES in young people with CP. To establish this, RSES was included in the test-retest in Study 2 (Chapter 5) and the exercise intervention in Study 3 (Chapter 7).

2.4.4 Summary

By reflecting on the OMs used in previous exercise studies, it is clear that there are many OMs available for use in assessing the effectiveness of

exercise programmes in young people with CP. As such, they provide information that can be used to guide clinicians regarding the outcome of rehabilitation programmes, including in the prescription of exercise for their patients.

For the purposes of Study 3 (Chapter 7), outcomes were chosen which would gather data on the 'body structures and function', 'activity', 'participation' and 'contextual factors' of the participants in accordance with the ICF model. OMs that had been evaluated for at least some of their psychometric properties were chosen to capture the wide range of areas that the proposed intervention sought to address.

Several systematic reviews of the psychometric properties of tools that measure different aspects of function in populations with CP have been published; for example, balance (Saether et al. 2013), quality of life (Carlon et al. 2010) and daily living activity (James et al. 2013). However, up to now there has been no systematic review conducted to look into the psychometric properties of OMs that assess gait quality and walking performance in young people with CP. Such a review would provide both the clinician and researcher with essential information regarding the reliability, validity and responsiveness of the measures used for gait quality and walking performance in young people with CP, and this would perhaps assist them in their choice of appropriate OMs to evaluate the management of their patients or the efficacy of an exercise intervention in research studies. Therefore, a systematic review of the psychometric properties of the measures used for gait and walking performance in young people with CP was undertaken for Study 1 (Chapter 4).

CHAPTER 3 GENERAL METHODOLOGY

3.1 Introduction

This chapter contains information regarding the methods and outcome measures used in Study 2 (test-retest reliability study, Chapter 5) and Study 3 (intervention study, Chapter 7). The protocols for both studies are described in their respective chapters (5 and 7). The psychometric properties of the main outcome measures have been discussed in the background chapter (Chapter 2), section 2.4.

3.2 Anthropometric measures

The anthropometric data measured includes body height (to the nearest centimetre) and body mass (to the nearest gram). Height was measured using a SECA height scale with the participant standing barefoot in the anatomical position against a wall. Body mass was measured using SECA scales with the participant barefoot in minimal clothing. Both of these measures were recorded for Studies 2 and 3. For Study 3, knee width (mm), ankle width (mm), leg length (mm) and tibial torsion (degrees) were also measured (Table 3.1). All of the above anthropometric measures were required for inputting into the Vicon Plug-In-Gait model. Regarding Study 2, no 3D motion analysis was performed; only body height and body mass were recorded for this study. Table 3.1 shows the details on how to measure knee, ankle width and tibial torsion.

Table 3.1 Details on the measurement of knee width, malleolar width, leg length and tibial torsion

Knee width	Measured between the medial and lateral epicondyles using large sliding callipers and applying gentle pressure.
Malleolar width	Measured between the medial and lateral malleoli using large sliding callipers and applying gentle pressure.
Leg length	When full knee extension is possible: Position the patient supine on the plinth with the pelvis as straight as possible. The trunk should be straight, head midline, arms by sides. The patient should not lift the head during measuring. Measured from the ASIS (press the end of the tape up against the underside of the ASIS) to the distal end of the medial malleolus. When significant knee deformity exists: Measured from ASIS to medial malleolus but via the medial condyle. Apply this method to both sides.
Tibial Torsion (transmalleolar axis)	Footprint method (Hazlewood et al. 2007): • lace the foot on a sheet of lined paper, with the knee axis parallel with the lines. The thigh should line up with long axis of the paper, with the tibial tubercle pointing forward. When the knee is flexed and extended, the foot should stay aligned with the long axis of the paper. • raw round the foot • ith a set square; mark a point vertically below the middle of each malleolus. • emove the paper and draw a line through the two points marking the malleoli–ankle axis. • easure the angle between the ankle axis and any of the lines on the sheet of paper; this is the angle of the transmalleolar axis. • xternal rotation of the foot (most common) is entered as a negative value in Vicon.

ASIS; anterior superior iliac spine

3.3 10m Shuttle run test (Studies 2 and 3)

Aerobic fitness was assessed using an adapted 10m shuttle run/walk test (10m SRT) as described by Verschuren et al. (2006). During the SRT participants walked/jogged/ran between two cones. They were instructed to

have arrived at the end of a ‘shuttle’ by the time the audio CD gave a bleep. For each level the time between the bleeps was reduced and the participants therefore had to increase their speed accordingly. If the participants were having trouble understanding the concept or adjusting their speed, the researcher ran the shuttles with them until they were comfortable with the concept. The 10m SRT was continued until the participants were unable to keep up with the pace of the bleeps, i.e. they failed to reach the cone before the next bleep. The participants were given the opportunity to ‘catch up’ on the next bleep if they failed to reach the end of one shuttle in time for the bleep. However, the test was ended if two consecutive shuttles were missed. The version of the 10m SRT used depended on the GMFCS level of the participant. Those classified at GMFCS levels I and II walked/ran between two markers set 10 metres apart; however, there was a different audio CD for the audio signals (‘bleeps’) for GMFCS levels I and II. The participants with GMFCS levels I and II started at speeds of 5km/hour and 2km/hour, respectively, with an increase of 0.25 km/h approximately every minute. Participants with GMFCS III used the same audio signals as those at GMFCS II; however, they walked/ran along the sides of a 7.5m square (thus requiring only a 90-degree turn and not a 180-degree turn at each bleep).

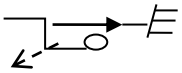
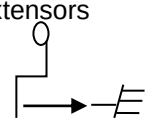
3.4 Strength test (Studies 2 and 3)

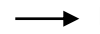
Isometric muscle strength (Nm) in a gravity-neutral position of the hip extensors, knee extensors and hip abductors was measured using a myometer (MIE ltd, Leeds, UK), according to Thompson et al. (2011). The test positions for each muscle group are described in Table 3.2. One padded strap was placed around the participant’s limb while the other strap was attached to a bar fixed to a wall (hip extensors, hip abductors) or plinth (knee extensors).

Three trials were carried out for each muscle group and the participants were given a 30-second rest between trials. The highest of the three trials was selected for analysis. The force values were multiplied by the lever arm to

derive moments and these were normalised by dividing by body mass. The lever arm was the distance from the estimated joint centre of rotation to the point of application of the force (i.e. placement of the strap), measured with a tape measure to the nearest mm (Table 3.2). The muscle groups were evaluated bilaterally.

Table 3.2 Details for isometric muscle testing

Muscle group	Position	Stabilisation	Resistance	**Lever arm** distance from force to
Hip extensors 	Supine – Hip at 90° knee at 90° with lower leg supported, low bench under lower leg (or held if this was not possible)	Hip belt – attached at an angle so it pulls downward and inferiorly	Distal femur, proximal to femoral condyles	Greater Trochanter
Hip abductors 	Supine – Knee propped up on blanket to allow comfortable flexion of hip and knee and to raise limb slightly	Hip belt	Distal femur, proximal to femoral condyles	Greater Trochanter
Knee extensors 	Seated knee at 90° on chair with backrest (back of chair is pushed up against the plinth so that it is stable)	Non-tested foot on floor	Distal Shank	Lateral knee joint line


Direction of resistance from the myometer
Direction of resistance from the hip belt for stabilisation

3.5 Three-dimensional gait analysis (Study 3)

Three-dimensional gait analysis (3DGA) with an eight-camera Vicon Nexus System (100Hz) (Oxford, UK) was used to record lower limb and pelvis kinematics. Participants had 14mm diameter passive reflective spherical markers placed on their lower limbs and pelvis in line with the Vicon Plug-In-Gait manual, which is based on the Helen Hayes marker system (Kadaba et al. 1990). A static trial was conducted using a Knee Alignment Device (KAD) to make a direct measure of the knee flexion extension axis. The KADs were

then removed and standard 14mm reflective markers were attached over the lateral epicondyle of each femur. Walking was assessed barefoot but with the participants' usual walking aids. The participants were asked to walk a distance of about six metres across the laboratory while the Vicon motion analysis system was recording. Between three and six gait cycles were recorded for each assessment. Prior to each gait analysis session, calibration of the VICON Workstation software was conducted, which involved two stages – static calibration and dynamic calibration. The calibration carried out strictly followed the manufacturer (VICON software) manual.

3.5.1 Data processing

The data were processed to enable further interpretation and analysis. The VICON Plug-In-Gait was used to derive joint kinematics and kinetics data. For the purpose of the current study (Study 3), only kinematics data were reported. Prior to processing the data, the anthropometric data of the participant (i.e. height (cm), body mass (kg), knee width (mm), ankle width (mm), leg length (mm) and tibial torsion (degrees)) were entered into the software. To process the data, one static (standing) trial and the appropriate walking trials were prepared for processing first. This involved several steps, the first of which was identifying the markers, which was conducted manually for the static trial. Once the static trial had been labelled, modelling for the static data was carried out by running a static gait model. Then, a single walking trial was labelled followed by detecting the gait events for one complete gait manually. This was then followed by running a dynamic gait model. The steps were repeated for each of the walking trials.

The processed data were then ready to be reported using the Polygon authoring tool. A report was created in the workstation under the respective session, which redirected the user to the Polygon authoring tool. The processed trials were then imported into the report and attached to the respective walks. The report includes graphical representation of the kinematics and kinetics of the data captured. The data were then exported to

Excel files and saved in their respective folders. The Excel files include numerical data for the temporal and spatial parameters, kinematics and kinetics for the movement recorded.

3.5.2 Gait Profile Score

The Gait Profile Score (GPS) is an index of overall gait pathology derived from specific gait parameters (can be taken over several kinematic variables along the entire gait cycle) and was calculated using the root-mean-square difference between a participant with CP data and the mean reference value of a healthy control in degrees (Baker 2009). GPS has previously been used to investigate improvements in gait quality as a result of single-event multi-level surgery in children with CP (Rutz et al. 2013) as well as to quantify change in gait quality following progressive resistance training in young people with CP (Taylor et al. 2013). A customised script was written and run in Matlab (Mathworks, Natick, MA) to determine the GPS from the Excel files derived from the Polygon authoring tool. The variables used to calculate the GPS were pelvic tilt, pelvic obliquity, pelvic rotation, hip flexion/extension, hip abduction/adduction, knee flexion/extension, ankle dorsiflexion/plantarflexion and foot progression angle in the transverse plane.

3.6 Gross Motor Function Measure 66 (Study 3)

Gross Motor Function Measure (GMFM-66) dimensions D and E were used to assess gross motor function. The test consists of 66 items that have been grouped into five different dimensions of gross motor function: dimension (A) lying and rolling; (B) sitting; (C) crawling and kneeling; (D) standing; and (E) walking, running and jumping. For this thesis (Study 3), only dimensions D and E were used to assess participants with CP. Each item is scored on a four-point scale, as follows: 0 = does not initiate; 1 = initiates; 2 = partially completes; and 3 = completes. For more detail on the scoring instructions for each of the items used, the reader is referred to Appendix 1. The overall score was derived from each category (dimensions D & E) for a participant and was divided by the total possible points to produce a category

percentage score. These percentages were averaged for both dimensions to yield an overall score. The overall score for the GMFM 66 was calculated using the Gross Motor Ability Estimator version 1 computer program.

3.7 Timed Up and Go test (Study 3)

The Timed Up and Go (TUG) test requires participants to stand from their sitting position on a standard chair with armrests (chair height 45 cm, as measured from the top of the seat to the floor), then walk 3m to a cone, turn around and sit down on the chair again. The time taken for this activity (from getting up from the chair, walking to sitting down again) was measured using a standardised timer (Digital stop-clock TM-20A) (Williams et al. 2005). The participants were instructed to walk at their normal walking pace and were permitted to use their normal walking aids. The participants conducted the test with their shoes on. Three trials were performed, with the average of the three trials used for analysis.

3.8 Objective habitual physical activity (Studies 2 and 3)

The objective habitual physical activity level was assessed using the ActivPAL^{TM} activity monitor (Grant et al. 2006) (PAL technologies Glasgow, UK). This activity monitor recorder is small in size (35 mm x 53 mm x 7 mm) and records the number of steps, sit-to-stand and stand-to-sit transitions and the time spent either walking, standing or sitting/lying over a 10-day period. This unobtrusive activity monitor is the size of a credit card and is attached to the front of the thigh using special double-sided adhesive pads (Palstickies^{TM}). The participants were asked to wear the monitor for 7 days but were allowed to remove it at night (sleep time) and when showering or swimming. In addition, the participants were asked to record the days and duration that the monitor was worn. The ActivPAL^{TM} data were transferred and processed using software (ActivPAL^{TM} version 6.4.1 PAL technologies Glasgow, UK), which classified the data into sitting (hours/day), standing (hours/day), stepping (hours/day), step counts (steps/day) and sit-to-stand

transitions (numbers/day). For analysis, the data were also saved as Excel files.

3.9 Short Form 12 version 2 (Study 2 and 3)

To measure the quality of life, the Short Form 12 (SF-12) version 2 was used which consists of 12 questions (Ware et al. 1996) (Appendix 4). An algorithm provided by the distributor was used to calculate the physical and mental component of the SF-12 scores. Healthy participants were required to complete the questionnaire during sessions 1 and 2.

3.10 Rosenberg Self-Esteem Scale (Studies 2 and 3)

The RSES is a questionnaire consisting of 10 statements with which participants can either strongly disagree, disagree, agree or strongly agree (Rosenberg 1965) (Appendix 5). The total score is derived from an algorithm, generating a score of between 0 and 30, with a higher value indicating higher self-esteem.

3.11 Canadian Occupational Performance Measure (Study 3)

The COPM was administered through a short interview with the participants to get them to identify those activities of daily living that they need to do, want to do or are expected to do (Law et al. 1998) (Appendix 3). At their first assessment, the participants were then asked to rate the importance of these activities and select the 5 most important. For the 5 most important measures, the participants were then asked to rate their level of performance and satisfaction on a 10-point scale. Performance was rated from 1: ‘not able to do it at all’ to 10: ‘able to do it extremely well’, and satisfaction was rated from 1: ‘not satisfied at all’ to 10: ‘extremely satisfied’. At the next assessments, the participants were reminded of the activities they had selected in the initial assessment, as well as their respective scores, and were asked to rate their performance and satisfaction on those activities again and also to rate their performance and satisfaction on those activities again. The participants were provided with their baseline score to provide

them with a reference for scoring the current score. Thus, if they felt they had improved, they could give a higher score, and vice versa. In pilot studies it was noticed that some people found it difficult to provide a performance score, hence the reason for providing them with their baseline score.

3.12 Gillette Functional Assessment Questionnaire (FAQ) (Study 3)

The FAQ is a self-reported questionnaire with a ten-level classification of walking and 22 functional mobility activities on a five-level Likert-type difficulty scale (Novacheck et al. 2000) (Appendix 2). The FAQ was designed for use in individuals with all levels of walking ability, and it focuses on what an individual can do independently with the use of assistive devices or orthoses, as needed to maximise function. The FAQ's 22 functional mobility activities were designed to provide further differentiation of higher ambulatory levels.

CHAPTER 4 PSYCHOMETRIC PROPERTIES OF MEASURES OF GAIT QUALITY AND WALKING PERFORMANCE IN YOUNG PEOPLE WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW

4.1 Introduction

The content of this chapter has been submitted for publication in Gait & Posture.

The transition from child to adult services is a challenging time for young people with CP because of their distinctive health, vocational and social needs and the inter-agency cooperation that this transition requires (Bakheit et al. 2009, National Institute for Health and Care Excellence 2016). A reduction in health services, such as physiotherapy, at this time point is reported in a range of countries worldwide (Wright et al. 2016, Binks et al. 2007). This is especially problematic as the research indicates that as young people with CP reach adulthood, they often experience decreased muscle strength accompanied by increased pain and joint deformity (Hilberink et al. 2007). Additionally, for many young adults with CP, walking ability often deteriorates (Bottos et al. 2001, Andersson and Mattsson 2001). Unsurprisingly, there is a growing interest in the identification of interventions that specifically address the gait deterioration seen among this age group (Shortland 2009). Crucial for this research, and indeed for any research into the efficacy of interventions, is the use of reliable, valid and responsive outcome measures, to meaningfully evaluate the success of interventions such as exercise programmes (Gabel et al. 2012). Psychometric properties have been defined as the elements that contribute to the statistical adequacy of a measurement instrument in terms of reliability, validity, measurement error and internal consistency (Gabel et al. 2002). Critically evaluating the

psychometric properties of available OMs can provide essential knowledge and evidence for clinicians and researchers, thereby allowing for the selection of the most appropriate OM(s) for a specific clinical or research question.

Three different research groups reviewed the measurement properties of measures of gait function and performance in neuro-paediatrics. The first review focused on the reliability and responsiveness of outcomes of gait function such as the Functional Mobility Score (FMS) and Gross Motor Function Measure (GMFM) (Ammann-Reiffer et al. 2014). Interestingly, in this review, little consideration was given to outcomes of gait quality (e.g. gait kinematics). Furthermore, these authors only reported on the measurement reliability and responsiveness, with no explicit consideration given to the validity of the OMs. The second review by Rathinam et al. (2014) critically appraised the reliability and validity of measures of gait quality but only those derived from observational gait assessment (OGA) tools used in paediatrics. Furthermore, the methodological quality of the outcome measures was not assessed using a standardised checklist such as the COnsensus-based Standards for the selection of health status Measurement INstruments (COSMIN) checklist (Terwee et al. 2012). As a result, it was not possible to assess the relative strength of the evidence provided to support their recommendations. A recent review appraised the OMs of walking ability in CP using the COSMIN checklist; however, this review focused only on OMs that were simple and quick to perform (Himuro et al. 2016).

The COSMIN checklist, which is recommended for use in systematic reviews of measurement properties (Terwee et al. 2012), has been used previously to explore the psychometric properties of gait function (Ammann-Reiffer et al. 2014) and other OMs such as balance, aerobic capacity and habitual physical activity in children with CP (Saether et al. 2013, Balemans et al. 2013, Mitchell et al. 2013). Considering the increasing number of studies reporting the results of gait analysis, ranging from visual observation scores

to computerised three-dimensional gait analysis (3DGA), a review of the psychometric properties of the outcomes of both gait quality (i.e. gait characteristics) and gait performance using a standardised quality checklist appears to be warranted. Therefore, the aim of this study was to evaluate the methodological quality and the strength of the evidence of studies that reported an evaluation of the psychometric properties of OMs of gait quality and walking performance in young people with CP. In doing so, the current study has restricted the review to include only those studies wherein the majority of the participants were of an age at which gait is likely to have matured and additionally included participants at such an age as when transition to adult services often takes places or is initiated.

4.2 Methods

4.2.1 Search strategies

The MEDLINE, CINAHL, PubMed and Scopus databases were searched using the main search categories of 'cerebral palsy', 'gait', 'outcome measure' and 'measurement properties' up to 14th January 2016. For the PubMed database, the same search strategy was applied but with a published additional sensitive search and exclusion filter for measurement properties (Terwee et al. 2009). Details pertaining to the search strategies employed are provided in Appendix 6. Finally, the reference lists of all the primary identified studies were manually searched and examined for studies that met the inclusion criteria.

4.2.2 Inclusion and exclusion criteria

The aim of this review was to explore and summarise the evidence presented in studies reporting the measurement properties of OMs for gait and walking performance in adolescents and young adults with CP just prior to and after transition to adult services. The current study therefore selected and implemented our inclusion criteria regarding age as follows: studies that 1) included one or more participants aged 16 years or over, as this is the age when transition from paediatric to adult services begins (Wright et al. 2016),

and 2) included participants with a mean age of ≥10 years, as this is the age at which gait has been shown to have matured (Berger et al. 1987). This review focused on five psychometric properties, namely reliability (test-retest reliability, inter-rater reliability and intra-rater reliability), measurement error, construct validity, criterion validity and responsiveness. The full list of study inclusion and exclusion criteria is shown in Table 4.1.

Table 4.1 Inclusion and exclusion criteria of the systematic review

Inclusion criteria
• Study population: children, adolescents & young adults with CP with at least one participant aged 16 years, mean age of participants in the study ≥10 years. • Studies reporting on psychometric properties (reliability, validity and responsiveness) of OMs of gait or walking performance. • The article should have been published in English. • Full-text, original article.
Exclusion criteria
• Abstracts, dissertation, conference proceedings, editorials, opinion pieces, review papers, letters, single case studies, short communications, technical notes. • Studies that validate translated versions of the OMs. • Primary aim of study is not to assess psychometric properties (e.g. intervention studies reporting on the practicality of the OMs). • Correlation between measures are investigated but the object of the study is not validation. • Studies that investigate the internal consistency of an OM.

CP: cerebral palsy; OM: outcome measure

4.2.3 Study selection process

After removing duplicates, two reviewers (AZ and MvdL) independently screened the titles and abstracts of the studies generated by the literature search based on the above-stated inclusion and exclusion criteria. In case of

disagreement or uncertainty, the full paper was reviewed. A third reviewer (KJ or TM) was available in the event that no consensus could be reached.

4.2.4 Quality assessment process

Full articles that met the inclusion criteria were independently rated by two reviewers (AZ as main and MvdL or KJ) using the COSMIN checklist. In the case of disagreement, there was a discussion to reach consensus. Each study was rated to determine (i) the overall methodological quality of the studies investigating specific psychometric properties, and (ii) the quality of the psychometric properties.

4.2.5 Evaluation of overall methodological quality scores

To determine the methodological quality of the studies, the COSMIN checklist was used (Terwee et al. 2012). The COSMIN checklist consists of nine boxes (internal consistency, reliability, measurement error, content validity, structural validity, construct validity, cross-structural validity, criterion validity and responsiveness), each comprising 5–18 items, for checking the methodological standards of the paper in terms of its design and statistical approach. Each item was scored on a four-point rating scale (‘poor’, ‘fair’, ‘good’ or ‘excellent’). The overall methodological quality score was based on the lowest rating of any items ticked in any box (Appendix 7).

The original COSMIN criteria, which were developed to assess the psychometric properties of self-reported questionnaires, and studies with a sample size of less than 30 were given a methodological rating of ‘poor’. It was anticipated that studies on the psychometric properties of gait quality and performance measures would often have less than 30 participants, and the application of the original criteria would exclude studies with otherwise good or excellent methodological quality. Consequently, in this review, the current study did not use the sample size item for the rating of any of the psychometric properties. Instead, sample size was accounted for at the best-evidence synthesis stage. This approach, which was first described by

Dobson et al. (2012) and was agreed by the COSMIN developers, was subsequently adopted in several other COSMIN reviews of outcomes used in the CP population (Ammann-Reiffer et al. 2014, Saether et al. 2013).

As the definitions and terminology of certain psychometric properties adopted by COSMIN may not always be the same as those used by the authors of the articles reviewed, the current study applied the COSMIN taxonomy as opposed to the terms used in the articles. As recommended by COSMIN, small modifications can be made to each scoring system to suit the purpose of the review or the characteristics of the outcome measures (Mokkink et al. 2010). The current study therefore developed 'rules' within the COSMIN rating to minimise the differences between reviewers in terms of their interpretations of the checklist items. The items relating to 'missing items' were not scored if the outcome measure was not a questionnaire or a test battery, as we regarded 'items' as questions in a questionnaire or parts of a test battery. With respect to the 'time interval appropriate' item, this was regarded as a time interval of two weeks or less to assess the test-retest reliability, as appropriate. For questionnaires, the minimum time interval for no recall was regarded as one week. Finally, in those studies assessing the validity of the observational gait analysis, 3DGA was accepted as the gold standard.

4.2.6 Evaluation of the quality of the psychometric properties

The quality of the psychometric properties, i.e. the strength of the evidence of the studies included in the review, was assessed using the quality criteria developed by Terwee et al. (2007), which have subsequently been revised by the author (Terwee 2012), as shown in Table 4.2. These guidelines were developed to score the quality of the studies in terms of their design, methods and outcome on the development and evaluation of the particular instruments. All of the OMs were rated as either 'positive' (+), 'indeterminate' (?) or 'negative' (-), depending on the results of the studies.

Of note, some of the OMs – for example the Edinburgh Visual Gait Score (EVGS) – may be given two ratings (i.e. 'positive' and 'negative'). This is because these OMs have more than one component for which the strength of the evidence can be rated; for example, 'knee angle at initial contact' and 'hip angle in stance'. If both a 'negative' and 'positive' rating are given for the different components, the evidence derived from these OMs will be rated as 'conflicting' in the synthesis of best evidence, as described in the next section.

Table 4.2 Quality measurements for psychometric properties (Terwee 2012 & Terwee et al. 2007)

Property	Rating	Quality criteria
Construct validity	+	At least 75% of the results are in accordance with these hypotheses
	?	No correlations with instrument(s) measuring related construct(s) AND no differences between relevant group reported
	-	Criteria for '+' not met
Criterion validity	+	Convincing argument that gold standard is 'gold' AND correlation with gold standard ≥0.70
	?	Not all information for '+' reported
	-	Criteria for '+' not met
Measurement error	+	SDC or LoA < MIC
	?	MIC not defined
	-	Criteria for '+' not met
Reliability	+	ICC or weighted Kappa ≥0.70
	?	Doubtful design or method
	-	ICC or weighted kappa <0.70, despite adequate design and method
Responsiveness	+	At least 75% of the results are in accordance with the hypotheses
	?	No correlations with changes in instrument(s) measuring related construct(s) AND no differences between changes in relevant groups reported
	-	Criteria for '+' not met

+ positive, ? Indeterminate, - negative
SDC: smallest detectable change, LoA: limit of agreement, MIC: minimal important change, ICC: interclass correlation coefficient

4.2.7 Synthesis of best evidence

A synthesis of best evidence for every psychometric property for each OM was constructed using the combined results of the methodological quality from the COSMIN checklist and the quality scores according to the adapted quality criteria (Terwee et al. 2007). The scores for these syntheses were adapted from the Cochrane Collaboration Back Review (van Tulder et al. 2003) and were also used in previous systematic reviews (Saether et al. 2013, Ammann-Reiffer et al. 2014). A general score was given to each OM and was either (i) strong – consistent findings in multiple studies of good methodological quality OR in one study of excellent quality; (ii) moderate – consistent findings in multiple studies of fair methodological quality OR in one study of good methodological quality; (iii) limited – one study of fair methodological quality; (iv) conflicting – conflicting findings; or (v) unknown – only studies of poor methodological quality or indeterminate evidence.

To account for the sample size, the following adapted ratings of the level of evidence were used: ‘strong’ when the total sample size of the combined studies was ≥100, ‘moderate’ for a total sample size between 50 and 99, ‘limited’ for a total sample size between 25 and 49 and ‘unknown’ when the sample size was fewer than 25 (Dobson et al. 2012).

4.3 Results

4.3.1 Descriptive

The search strategy resulted in the identification of 3318 articles. Figure 4.1 shows the selection process used for the articles included in this review. Initial screenings based on the title resulted in the exclusion of 3089 articles. A further 45 and 69 articles were omitted based on the reviews of abstracts and full text, respectively. A final number of 20 articles reporting on 14 OMs were included for review via the COSMIN checklist, quality rating and synthesis of best evidence. The 14 OMs included in this review are shown in

Table 4.3. In agreement with the inclusion criteria, some studies were excluded from the present review because the study population did not fit the inclusion criteria or because the OMs of gait quality and walking performance were not the primary constructs of the outcome measure. In these instances, they were part of a wider assessment of physical and motor function, such as the Functional Independence Measure (WeeFim) and GMFM.

The average age in the studies included for this review ranged from 10.3 to 14.97 years. In terms of sample size, only 5 out of the 20 studies included less than 30 participants. Reliability was assessed 17 times (inter-rater reliability n=10, intra-rater reliability n=2 and test-retest reliability n=4) and measurement error once. For eleven OMs the validity was assessed 16 times (construct n=13 and criterion n=3). Responsiveness was assessed only four times in four different OMs.

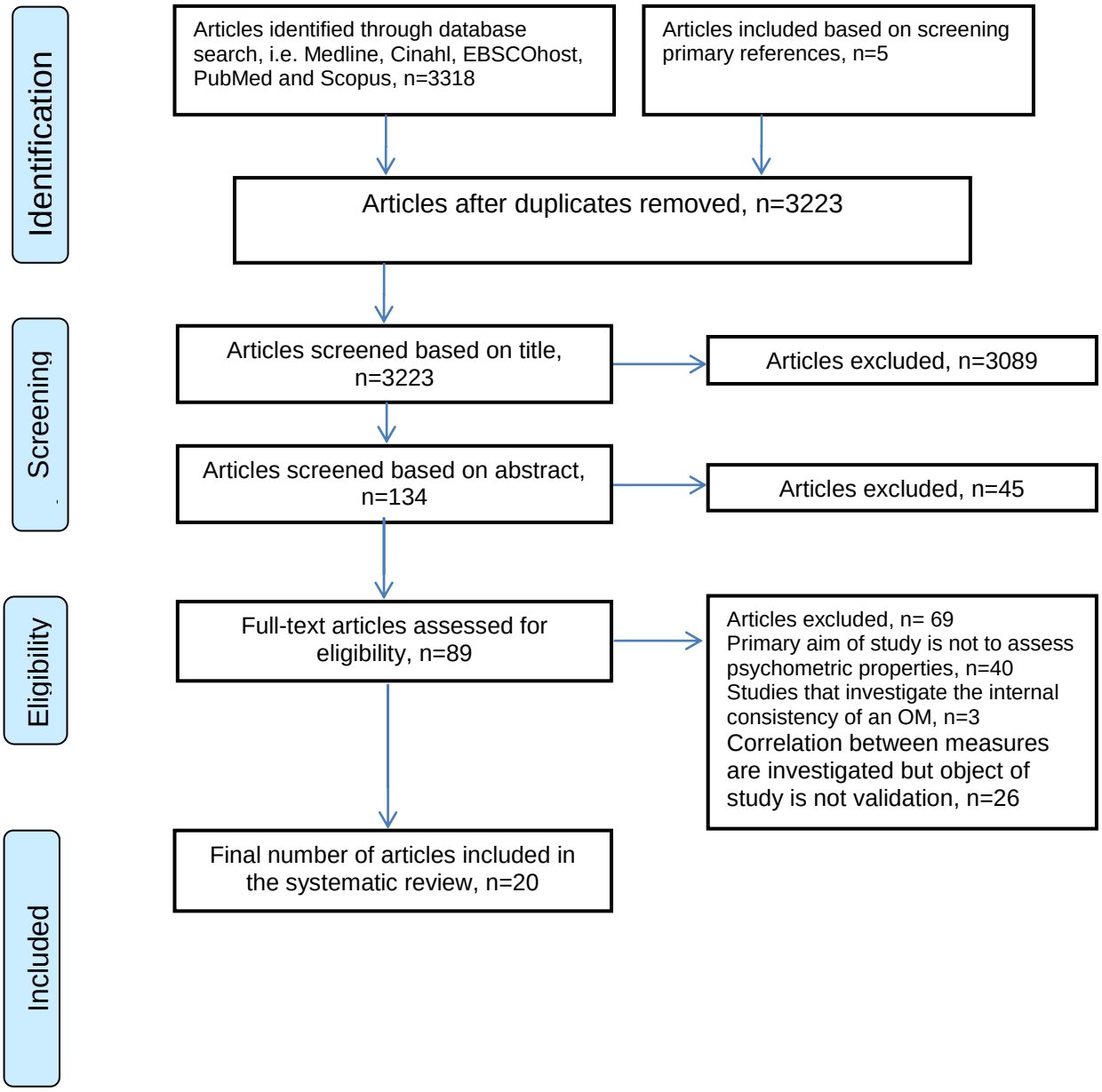


Figure 4.1 Flow chart of the selection process of articles included in the review

Table 4.3 Characteristics of the studies and ratings of methodological quality and quality of evidence of the included studies

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
Visual Gait Assessment Scale (VGAS)	Dickens 2006	CP,31,5–17 (mean 10.6)	Intra-rater reliability	Kappa Observer 1: -0.04–0.76, 95% CI (-0.43–1.00) Kappa Observer 2: (0.07–0.84), 95% CI (-0.32–1.00)	Good	+ knee peak extension in TS,IFC, foot contact in stance, timing of heel rise, knee flex in sw,foot in stance) -hip in TS &swing, kneeflex in sw),IFC
			Inter-rater reliability	Kappa Session 1: (0.44–0.79), 95% CI (0.17–0.96) Kappa Session 2: (0.05–0.75), 95% CI (0.05–1.00)	Good	+ foot contact in stance, initial foot contact - both observer hip in TS, MS, knee in TS & swing, timing heel rise, knee peak extension
			Criterion validity	Kappa VGAS & 3DGA: min KF -0.11 (95% CI -0.38–0.16) - max KE 0.51 (95% CI 0.26–0.76)	Excellent	-
Visual gait score (VGS)	Kawamura et al. 2012	CP,50,>8 (mean 13.7)	Inter-rater reliability	Kappa Lt HA at LR 0.25 - Rt IFC 0.88 (95% CI was not reported)	Fair	+ ankle DF in IC - HE in TS,KF in IC, KE in TSt, KF in Sw, pelvic obliquity, Had in LR, pelvic rotation, hip rotation in MSt, foot progression at Mst

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
			Criterion validity	VGS & 3DGA Kappa IFC 0.01 – KF at IC 0.65) 95% CI was not reported)	Good	-
Edinburgh Visual Gait Score	Viehweger 2010	CP,10,9–16 (mean 12.6)	Intra-rater reliability	ICC: -0.34 to 0.99 (95% CI was not reported)	Good	+ for total score (OS,OS+GLM,NS, resident OS, PT+GLM), for trunc (resident OS), PT+GLM, for hip (OS+GLM, PT+GLM), for knee (all observer except P+GLM, PT), for foot (all observer except PT) - for total (P+GLM,PT), for trunc (all observer except resident OS+GLM), for pelvis (all observers), hip(all observers except OS+GLM, PT+GLM),knee (P+GLM, PT), foot (PT)
			Inter-rater reliability	% agreement Max KE 21.4% - Foot rotation & max lateral trunk shift in stance	Good	?

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
				67.9		
	Gupta 2012	CP,50,6–19 (mean 11.44)	Responsiveness	MCID After 6mo 10.62, 12mo 14.98 Effect size After 6mo 1.19, 12mo 1.22	Poor	?
Physician rating scale	Wright 1999	CP,25,6–18 (mean 11.2)	Test-retest reliability	Within session ICC: 0.65-0.93 (95% was not reported) Between session ICC: 0.45 0.94 (95% was not reported)	Good	+ for (step length, foot angle, velocity) - for (step width)
			Inter-rater reliability	ICC: 0.84-0.99 (95% CI was not reported)	Good	+
Timed up & down stairs (TUDS)	Chrysagis et al. 2014	CP, 35,12–18 (mean 14.97)	Inter-rater reliability	ICC: >0.90 (95% CI was not reported)	Fair	+
			Construct validity	TUDS & GMFM88 r= -0.5, p<0.01	Fair	+
Timed Up Go Test (TUG)	Chrysagis et al. 2014	CP, 35, 12–18 (mean 14.97)	Inter-rater reliability	ICC: >0.90 (95% CI was not reported)	Fair	+
			Construct validity	TUG & GMFM88 r= -0.6, p<0.01	Fair	+
	Hassani et al.	CP,168, 8–18 (mean	Construct validity	ANOVA:	Fair	+

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
	2014	12.9)		GMFCS I<III, GMFCS II<IIII, p<0.01		
			Responsiveness	Mean change GMFCS I,II,III non - surgical /surgical: 0.4s,0.2s,-0.1s/–0.1s,0.6s,3.0s	Poor	?
1 minute walk test (1MWT)	McDowell 2009	CP,34, 4–16 (mean 11.3)	Construct validity	1MWT (distance) & GMFM88 scores r^2=0.84, p<0.001 1MWT (distance) & GMFM66 scores r^2=0.80, p<0.001	Fair	+
	Kerr et al. 2007	CP,46, 5–17 (mean 11.68)	Construct validity	1MWT (distance) & 0_2 cost r=0.69p<0.001	Good	+
	Chrysagis et al. 2014	CP, 35, 12–18 (mean 14.97)	Inter-rater reliability	ICC: >0.90 (95% CI was not reported)	Fair	+
			Construct validity	1MWT & GMFM88 r=0.8, p<0.01	Fair	+
	Hassani et al. 2014	CP,168, 8–18 (mean 12.9)	Construct validity	ANOVA: GMFCS I>II, GMFCS I>III, GMFCS	Fair	+

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
				II>III, p<0.01		
			Responsiveness	Mean change GMFCS I,II,III non-surgical/surgical: 1.1,4.4,4.3/10.7.5.6.-4.9	Poor	?
Functional Mobility Scale (FMS)	Harvey et al. 2007	CP,118,2–18 (mean 10.3)	Inter-rater reliability	Kappa; agreement: 5m 0.87, 96%, 50m 0.92, 98%,500m 0.86, 96%	Good	+
	Harvey et al. 2010a	CP,66,6–16 (mean 10.0))	Responsiveness	OR(CI) 3mo, 6mo,24mo 5m: 0.13(0.07–0.24), 0.36(0.23–0.58), 2.08(1.33–3.24) 50m: 0.09(0.04–0.17), 0.32(0.19–6.55), 2.16(1.37–3.41) 500m: 0.24(0.14–0.43), 0.50(0.32–0.8), 2.23(1.44–3.45)	Fair	?
	Graham et al. 2004	CP,18,8–17 (mean 12.8)	Construct validity	FMS & observation Weighted kappa: 5m=0.71, 50m=0.76, 500m=0.74	Good	+
	Chrysagis et al.	CP, 310,4–18 (mean	Inter-rater reliability	ICC: 5m 0.95, 50 m 0.94, 500 m	Good	+

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
	2014	11.0)		0.95 (95% CI was not reported)		
10 metre Fast Walk test	Chrysagis et al. 2014	CP, 35, 12–18 (mean 14.97)	Inter-rater reliability	ICC: >0.90 (95% CI was not reported)	Fair	+
			Construct validity	10MWT & GMFM88 r= 0.6, p<0.01	Fair	+
6 minute walk test (6MWT)	Nsenga Leunkeu et al. 2012	CP,41,11–16 (mean 13.6)	Test-retest reliability	ICC (single session, 30mins interval) 0.93–0.98 (95% CI was not reported)	Fair	+
			Measurement error	LOA: -44m and 42m	Fair	?
	Bagley 2007	CP,12,10–16 (mean 14.2)	Test-retest reliability	ICC (one week) 0.8 (95% CI was not reported)	Good	+
			Construct validity	6MWT(distance) & VO_{2peak} r=0.625, p<0.05	Fair	+
Gillette functional Assessment Questionnaire (FAQ)	Mackey 2005	CP, 758,4–18 (mean 11.0)	Construct validity	GMFCS I & II – GMFM66 AUC 90.3%, sensitivity 86.6%, specificity 82.8%, p<0.001 GMFCS II & III GMFM66-E AUC	Fair	+

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
				94%, sensitivity 93.3%, specificity 82.2%, p<0.001		
3D gait – kinematics, temporal spatial	Massaad 2014	CP,10,5–16 (mean 12.0)	Test-retest reliability	Within session (r) Sagittal: tr tilt 0.82 – ankle DF 0.98 Frontal: pelv obliquity 0.88 – tr obliquity 0.91, p<0.001 Transverse: tr rotation 0.81 – foot rotation 0.91, p<0.01 Between session Sagittal: pelvic tilt 0.93 – KF/KE 0.99 Frontal: foot progression 0.81 – HA, HAb 0.95, p<0.01 Transverse: Kn rotation 0.82 – foot rotation 0.92, p<0.01	Good	+
GDI	Molloy 2010	CP,134, 5–20 (mean 10.5)	Construct validity	GDI & GMFCS r= -0.44, p<0.01	Poor	+

Outcome measure	Study	Diagnoses, n, age (y)	COSMIN measurement property	Results	Rating	
					Methodological quality (modified)	Quality of evidence[b]
	Baker 2009	CP & healthy,150(CP),4–17 (mean 10.8)	Construct validity	GDI & GMFM66 r=0.70, p<0.001 GDI & GMFM88 r= 0.67, p<0.001	Fair	+
GPS	Massaad 2014	CP, Orthopaedic& neurology condition, 271 (CP), <18 (mean 12.0)	Criterion validity	GPS & GDI r=0.995, p<0.001	Good	+
	Baker et al. 2009	CP, Orthopedic & neurology condition, 271 (CP), <18 (mean 12.0)	Criterion validity	GPS & GDI r=0.995, p<0.001	Good	+

GMFCS-Gross motor function classification score, SEM-standard measurement error, LOA-limit of agreement, OR-odds ratio, AUC-area under the curve,CI-confidence interval, SD-standard deviation, MDC-minimal detectable change, MCID-minimal clinically importance difference, m-metres, mo-months, s-seconds, ICC-interclass correlation coefficient, CV-coefficient of variance, r-Pearson correlation, pelv-pelvic, tr-trunk, KF-knee flexion, KE-knee extension, DF-dorsiflexion, MS-midstance, MSw-Mid swing, TS-terminal swing, IFC-initial foot contact, KF-knee flexion, KE-knee extension, HA-hip adductor, HAb-Hip abduction, HF-hip flexion, HE-hip extension, IR, internal rotation, LR-loading response, Rt-right, Lt-left, pelv-pelvic, tr-trunk, Max-maximum, Kn-knee, GVS- gait variables score, GPS- gait profile scores, GDI- gait deviation index, OS-orthopedic surgeon, GLM-gait lab member, NS-neurosurgeon, PT-physiotherapist, P-physiatrist, [b]Terwee et al. 2007

4.3.2 Overall methodological quality scores

The methodological quality of all the studies included in this review was rated using the modified COSMIN checklist, and the results are shown in Table 4.3. The methodological quality of the studies assessing the psychometric properties of the OMs was rated as ‘excellent’ (once), ‘good’ (14 times), ‘fair’ (18 times) and ‘poor’ (four times). The most common reason for a ‘poor’ rating was inappropriate statistical analysis used in the studies.

4.3.3 The quality criteria of the psychometric properties (Strength of evidence)

In terms of quality criteria, the majority of the psychometric properties of the OMs in this review were rated as ‘positive’ (24 times), with the remainder being rated as ‘indeterminate’ (six times), ‘negative’ (twice) and ‘positive/negative’ (five times). The inter-rater, intra-rater and test-retest reliability of OMs such as Visual Gait Assessment Scale (VGAS), Visual Gait Score (VGS), EVGS and Physician Rating Scale (PRS) were rated both ‘positive/negative’, as these OMs included the rating of different gait events (e.g. initial contact, heel strike, etc.) or gait characteristics (i.e. peak knee flexion in swing). For example, the intra-rater and inter-rater reliability of VGAS, VGS, EVGS and PRS all received a ‘positive’ rating for type of initial foot contact. In contrast, the inter-rater reliability of VGAS and VGS to determine hip position in terminal stance and knee flexion in swing was scored as ‘negative’ (Dickens 2006, Kawamura et al. 2007).

4.3.4 Synthesis of best evidence

The overall levels of evidence, synthesising methodological quality and the strength of the evidence for each of the OMs, are presented in Table 4.4. Only the scores on the FMS were found to have ‘strong’ levels of evidence regarding inter-rater reliability. ‘Moderate’ levels of evidence were found for criterion validity of the Gait Profile Score (GPS). The majority of the OMs have limited positive evidence for one or more psychometric properties, i.e.

Timed up and down stairs (TUDS) (inter-rater reliability, construct validity), TUG (inter-rater reliability, construct validity), 1MWT (inter-rater reliability, construct validity), 10m FWT (inter-rater reliability, construct validity), 6MWT (test-retest reliability), FAQ (construct validity) and GDI (construct validity). The evidence for the reliability of VGAS, VGS and PRS was found to be 'conflicting'. The evidence for the responsiveness of all four of the OMs for which this was investigated was rated as 'unknown'.

Table 4.4 Synthesis of quality rating for each outcome measure

Domain							
	Reliability				Validity		
OMs	**Test-retest**	**Intra-rater**	**Inter-rater**	**Measurement error**	**Construct**	**Criterion**	**Responsiveness**
VGAS		+/-	+/-			-	
VGS			+/-			-	
EVGS		?	?				?
PRS	+/-		+/-				
TUDS			+		+		
TUG			+		+		?
1MWT			+		+		?
FMS			+++		?		?
10m FWT			+		+		
6MWT	+			?	?		
FAQ					+		

	Domain						
	Reliability				Validity		
OMs	**Test-retest**	**Intra-rater**	**Inter-rater**	**Measurement error**	**Construct**	**Criterion**	**Responsiveness**
3DGA	?						
GDI					+		
GPS						++	

+++ or ---strong: consistent findings in multiple studies of good methodological quality OR in one study of excellent quality
++ or – moderate: consistent findings in multiple studies of fair methodological quality OR in one study of good methodological quality
+ or – limited: one study of fair methodological quality
+/- conflicting: conflicting findings
? unknown evidence: only studies of poor methodological quality and/or indeterminate rating for measurement property
VGAS-visual gait assessment scale, VGS-visual gait score, EVGS-Edinburgh Visual Gait Score, PRS-Physician Rating Scale, TUDS-Timed up and down stairs, TUG-Timed up and go test, 1MWT-one minute walk test, FMS-functional mobility scale, 6MWT-six-minute walk test, FAQ-Gillette functional assessment questionnaire, 3DGA-three-dimensional gait analysis, GDI- gait deviation index, GPS-gait profile score

4.4 Discussion

This is the first systematic review of studies evaluating the psychometric properties of both gait quality and walking performance OMs in young people with CP using standardised checklists to assess both methodological quality and strength of evidence.

The current study identified 14 OMs, the psychometric properties of which were studied, that were used to assess gait quality and walking performance, with a CP population which included at least one person aged 16 or over and with an average age of 10 years and over. Of these 14 OMs, none had a strong level of evidence for all of the psychometric properties considered in this review. Only one measure (FMS) was found to have a strong level of evidence regarding reliability. The reason for this may be attributed to the relatively simple rating system used in the FMS. Our finding of strong evidence in the reliability of FMS was in agreement with Ammann-Reiffer et al. (2014), who used the same methodology in their synthesis of the level of evidence. While there was strong evidence as to the reliability of FMS, the evidence for the construct validity and responsiveness of FMS was rated 'unknown' in this review. The level of evidence with regard to the reliability of 6MWT and TUG was rated as 'limited' in the current review, while in contrast, Ammann-Reiffer et al. (2014) reported a moderate level of evidence of reliability for both measures. The discrepancy between the results is likely due to the different number of studies included for review, since Ammann-Reiffer et al. (2014) included all studies with participants between 1 and 18 years old.

Although the current review showed limited evidence for the reliability and none for the responsiveness of the 6MWT and TUG, these OMs have been used in the CP population to assess the effect of exercise interventions (Taylor et al. 2012, McNee et al. 2009). The likely reason for this use is that both OMs are very simple, practical and easy to perform. However, both TUG and 6MWT provide information only on gait performance and not gait quality

such as gait kinematics. The latter information is obtained via both instrumented and observational gait analysis and is also of interest when monitoring potential gait deterioration in early adulthood (Shortland 2009).

A previous review of five OGA measures suggested EVGS as being the best currently available OGA with which to assess gait pattern in children with CP (Rathinam et al. 2014). Interestingly, this recommendation did not seem to be based on methodological ratings of reliability and validity, as EVGS did not obtain higher scores compared to other OGAs in that review. In addition, the rating appraisal used in this previous review focused primarily on methodological qualities with no explicit rating for the strength of evidence, as in the guidelines proposed by Terwee et al. (2007) which were applied in the current review. Comparison with our results is therefore difficult.

The majority of the visual observation scores received 'conflicting' evidence for their reliability because the ratings for several components of the score were reliable while others were not. The limited positive evidence for the reliability of gait kinematics was reduced to 'unknown' because of the small sample size of the only study (Mackey 2005) in our review which reported on this psychometric property. The evidence for the construct validity of the GDI was 'limited' because of the 'fair' and 'poor' methodologies of the two studies which reported on this outcome measure (Massaad 2014, Molloy 2010).

The methodological quality of the majority of the psychometric property studies in this review was rated as follows: 'excellent' (once), 'good' (14 times), with approximately one-third rated as 'fair' (18 times) or 'poor' (four times). The majority of the studies displayed similar methodological shortcomings. For reliability studies, the common reason for not being rated 'excellent' was that the model of the ICC was not reported.

One study (Harvey et al. 2010b) that employed ordinal scoring did not report the weighted kappa coefficient, thus reducing its rating to 'good'. In terms of

studies assessing construct and criterion validity, no hypotheses were formulated in the majority of the studies (Chrysagis et al. 2014, Hassani et al. 2014, Bagley 2007, Molloy 2010) and this resulted in a ‘fair’ rating.

Responsiveness was reported only for the FMS, EVGS, TUG and 1MWT in three separate studies (Gupta 2012, Hassani et al. 2014, Harvey et al. 2007). However, for all of these OMs the evidence-level rating for responsiveness was classed as ‘unknown’. The reason for this lack of evidence was due to the ‘poor’ rating of the methodological quality for EVGS (Bagley 2007), TUG and 1MWT (Hassani et al. 2014) and the ‘indeterminate’ rating for FMS in terms of quality criteria (Gupta 2012).

This review also highlights the limited number of studies that included participants aged 16 and above and where the majority were of an age at which gait had matured (Berger et al. 1987). Applying our inclusion criteria regarding age resulted in the omission of 30 studies, thereby leaving only 14 studies available for this review. The majority of the studies omitted included much younger participants. This may be expected as CP is often considered as a childhood disease and the majority of interventions, such as multi-level surgery and botulinum toxin injections, take place in childhood.

Young people affected by CP aged 16 years and older are often not included in intervention and psychometric studies. This is problematic as this is the age group at which both the transition from paediatric to adult health services is initiated or occurs (Wright et al. 2016, Binks et al. 2007) and gait function often starts to deteriorate (Bottos et al. 2001, Andersson and Mattsson 2001). Future studies should therefore evaluate interventions addressing the gait deterioration that often occurs during adolescence or early adulthood while others should assess the psychometric properties of the OMs commonly used with this age group.

Although we did not score the sample size items in the COSMIN checklist, we did account for sample size in our synthesis of best evidence after combining the sample sizes of studies with a similar methodology, as carried out in previous reviews (Ammann-Reiffer et al. 2014, Dobson et al. 2012), in agreement with the COSMIN developers. Even after adding the sample sizes of those studies with the same methodology, the psychometric properties of the OMs did not reach an adequate level of evidence due to the small sample size of the individual studies. Future research on the psychometric properties of the measurements to be used in populations of young people with CP should consider adopting multi-centre research designs that could overcome this issue of sample size.

This review has a few other limitations. Firstly, the current study did not include composite OMs that have walking performance as part of the assessment, such as the GMFM and WeeFim, because these OMs have been included in other reviews on activity limitation and functional motor abilities in children with CP (Harvey et al. 2008, Debuse and Brace 2011, Ketelaar et al. 1998). Another limitation of this review is the language restriction, since we opted to include only English-language articles.

Additionally, while the current study acknowledges the importance of the clinical utility of any OMs, this review focused only on the psychometric properties of the OMs of gait quality and walking performance. 3DGA is likely to be impractical in most clinical settings due to the increased time, cost and space requirements associated with this type of assessment. In comparison, the FMS is a very simple questionnaire used to describe functional mobility and could potentially be routinely implemented in clinical practice to assess walking performance in young people at their transition from paediatric to adult services.

4.5 Conclusions

This systematic review reported the psychometric properties of OMs assessing gait quality and walking performance in young people with CP. Only the reliability of FMS was found to have a ‘strong’ level of evidence, with the majority of the outcome measures having a conflicting or limited level of evidence. No evidence was found for responsiveness for any OMs in this review. This result is of concern as any ability to assess changes in gait quality and/or walking performance in patients following an (exercise) intervention requires OMs that are responsive to change. Future research is suggested to provide more studies with a high-quality design to assess the responsiveness of OMs of gait quality and walking performance in young people with CP, especially of those who are just coming up to and following transition to adult physiotherapy services.

CHAPTER 5 RELIABILITY OF PHYSICAL FUNCTION, HABITUAL PHYSICAL ACTIVITY, QUALITY OF LIFE AND SELF-ESTEEM OUTCOME MEASURES IN YOUNG PEOPLE WITH CEREBRAL PALSY AND AGE-MATCHED CONTROLS

5.1 Introduction

Assessing physical function, habitual physical activity, quality of life and self-esteem using valid and reliable outcome measures (OMs) is important to evaluate the effects of treatment/interventions for individuals with CP. One essential psychometric property of an OM is reliability, which refers to the reproducibility of measurements (Portney and Watkins 2000) or, more specifically, the extent to which scores for patients who have not changed are the same across repeated measurement (Mokkink et al. 2012).

An OM's reliability is assessed by administering a test to a group of people, in the same way, on two or more different occasions, hours, days or weeks apart (Portney 2000, Mokkink et al. 2012). Test-retest reliability assures clinicians and researchers that the OM yields the same result, in a stable patient or participant, each time it is used.

According to Nelson et al. (1979), two assumptions need to be considered in test-retest reliability assessment. Firstly, the true score does not change between administrations. Secondly, the time period between administrations is sufficiently adequate to prevent learning, carry-over effects or recall (Nelson et al. 1979). An understanding of the stability or variability in the outcome being measured, and the characteristics of the participants involved in the reliability study, should guide the time interval between administrations (Vaz et al. 2013).

The reliability of physical function OMs such as the Timed up and Go test (TUG), 10m Shuttle Run Test (SRT) and isometric muscle strength (ISM), habitual physical activity (activity monitor), quality of life measured using Short Form 12 version 2 (SF-12) and the RSES has been reported previously and is discussed in Chapter 2 (Section 2.4). However, there is a lack of evidence concerning the reliability, in particular the test-retest reliability, of these OMs in young people with CP, especially in those just before and after the transition to adult physiotherapy services, i.e. those aged 16 and over.

5.2 Study aims

- To establish the test-retest reliability of physical function OMs (TUG, SRT, ISM), habitual physical activity (activity monitor), quality of life (SF-12) and self-esteem (RSES) in young people with CP.

- To establish the test-retest reliability of physical function OMs (TUG, SRT, ISM), habitual physical activity (activity monitor), quality of life (SF-12) and self-esteem (RSES) in age-matched healthy controls.

5.3 Methods

5.3.1 Participants

For the CP group, the participants were identified from the patient database of the Anderson Gait Analysis Laboratory, SMART centre, Astley Ainslie, Edinburgh. The inclusion criteria for the CP group were (a) individuals with CP between 16 and 25 years of age, (2) able to ambulate 100 metres with or without aids. Participants or the carer (on behalf of the participant) were asked to fill in the questionnaires (SF-12 and RSES) before the baseline assessment took place. Patients were excluded from the study if they had any of the following: (a) insufficient cognitive ability to give informed consent, understand and provide answers to the questions in the questionnaire

booklet, (b) medical contraindications to participate in exercise testing (American College of Sports Medicine 1991), or (c) orthopaedic surgery or Botox injection in the last 6 months. Participants with CP for the current reliability study comprised a subset of participants (those assigned to the control group) of the RCT (into the effects of a community exercise programme) (Study 3, Chapter 7).

For the age-matched healthy controls, a convenient sample of 18 participants was recruited from Queen Margaret University (QMU) through online advertisement in the QMU moderator and through the local managers of rugby clubs near Musselburgh and Edinburgh, with ages ranging between 16 and 25 years. The justification for assessing the test-retest in healthy controls was twofold: firstly, to be able to compare the test-retest reliability between the two populations, thus providing more insight into the consistency and measurement error of these OMs. Secondly, the test-retest reliability assessments of the healthy controls allowed for more practice with the OMs prior to conducting the assessments with the primary population, i.e. young people with CP.

Participants were excluded if they had any neuromuscular, musculoskeletal or cardiopulmonary disease which would affect their physical function. This study was approved by the QMU Research Ethics Board and National Health Services (NHS) research ethics committee (see Appendix 8).

5.3.2 Study protocol

All study assessments took place in the Motion analysis Lab at Queen Margaret University (QMU), Edinburgh except for those for the 10m SRT, which took place at the sports hall of the QMU sport centre. Participants gave their informed consent prior to participating (Appendix 9) in the baseline measurement (T0). After informed consent was given by the participants, the assessment started with the TUG and ended with the 10m SRT (Figure 5.1).

Basic demographic data for the age, date of birth, date and time of assessment, with the GMFCS level also recorded for the CP group, was recorded using a standardised data collection sheet (Appendix 10). After completion of the assessments, the activity monitor (ActivPAL^{TM}) and adhesives to attach the activity monitor to the leg (Palstickies^{TM}) were given to the participants in addition to verbal and written instructions regarding usage of the activity monitor usage (Appendix 11). Stamped addressed envelopes were provided to the participants for returning the ActivPAL^{TM} after seven days, where appropriate (i.e. only for those participants who were non-QMU based). Otherwise, the participants were instructed to drop off the ActivPAL^{TM} at the QMU school office. Session 2 (T1) was planned for approximately 6 weeks after T0, whereby all of the OMs were repeated, and this was a period during which no changes in the measures of physical function, habitual physical activity, quality of life and self-esteem were expected. The order of the measurements was exactly the same for both sessions.

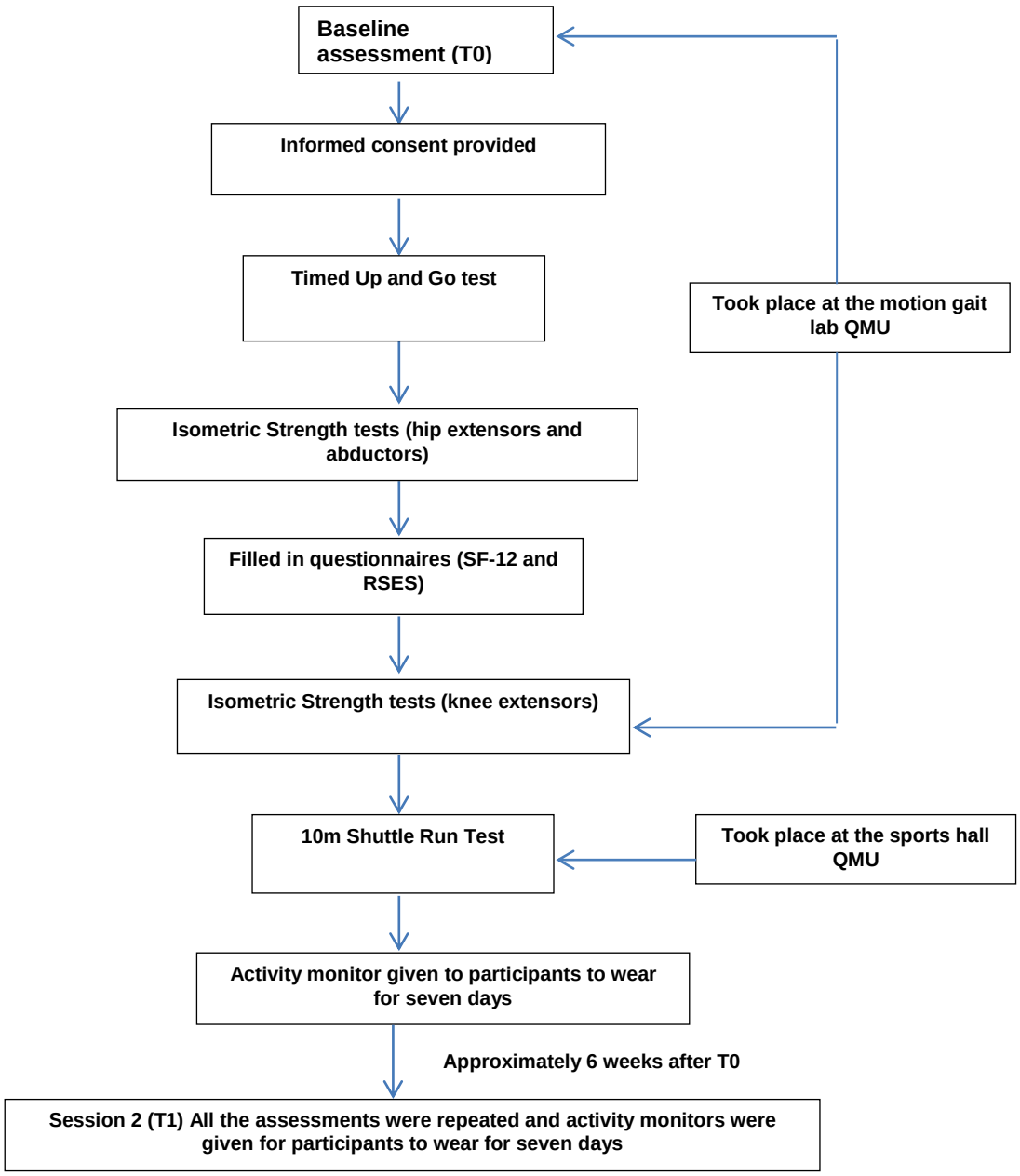


Figure 5.1 Flow chart during the baseline assessment (T0) and session 2 (T1) for both CP group and healthy controls

5.3.3 Outcome measures

Three tests (TUG, isometric strength tests, 10m SRT) and two questionnaires (SF-12 v2, RSES) were administered twice, with approximately 6 weeks between the assessments. In addition, the participants were required to wear the activity monitor (ActivPAL^{TM}) for seven days. For more details on the outcome measures, readers are referred to the general methodological chapter (Chapter 3).

5.3.4 Statistical analysis

Statistical analysis was performed using Statistical Package for Social Science (SPSS) version 21. Reliability (consistency) was assessed using the ICC, including 95% confidence intervals (CIs). The ICC model (2,1) was used to generalise the reliability of the data beyond the confines of the current study. Furthermore, the ICC model (2,1) used in the current study takes account of variance between the test-retest scores and was applicable because each participant was measured on each occasion (Portney 2000). ICC values >0.75 indicate good reliability, 0.50 to 0.75 indicates moderate test-retest reliability and <0.50 represents poor test-retest reliability (Portney 2000).

In addition, standard error of measurement (SEM) and minimal detectable change (MDC) were calculated to quantify the measurement error in the units of measurement used in this study. The SEM determines how much a score varies with repeated measures, taking into account the within-patient variability of the score (Harvill 1991, Weir 2005). Derived from the SEM is the MDC, which is considered to be the minimal amount of change that is not likely to be due to chance variation in the measurement. The MDC thus provides information on the minimal difference needed to consider the change as being a 'true' change. The SEM was calculated as SD x $\sqrt{1-ICC_{(2,1)}}$, where SD is the standard deviation of the grand mean (mean of session 1 and session 2) from all participants (Flansbjer et al. 2005). The MDC was calculated at a 95% confidence level as SEM x 1.96 x $\sqrt{2}$. The MDC is presented both in the unit of measurement and as a percentage of the grand mean (MDC%). MDC% was calculated by dividing the MDC by the average of the measurement (both sessions) and multiplying by 100.

The participants' habitual physical activity data derived from the ActivPAL^{TM}, were included for analysis if at least four full days (24 hours each) of data were available. The data were visually inspected and from this it was subjectively judged whether the ActivPAL^{TM} was worn or not.

5.4 Results

5.4.1 The test-retest reliability of the physical function outcome measures (TUG, SRT, ISM), habitual physical activity (activity monitor), quality of life (SF-12) and self-esteem (RSES) in young people with CP

The overall mean age for the CP participants for this reliability study was 20.28 (3.43) years. Test-retest reliability data were available for eight CP participants (n=5 females) with GMFCS I–III (Table 5.1). From Table 5.1, the participants with GMFCS level I have the highest mean body mass, at 90 kg, in comparison with the participants with GMFCS levels II and III, who have a mean body mass of 55.0 kg and 49.5 kg, respectively. The two participants with CP classified as GMFCS level I had the highest mean body mass index (BMI=26.4kg/m^2) compared to the participants with CP classified as levels II and III.

Table 5.1 Characteristics of participants with cerebral palsy (n=8)

	Overall	GMFCS I n=2	GMFCS II n=3	GMFCS n=3
Gender (female:male)	5:3	0:2	3:0	2:1
Mean Age (SD) (yrs)	20.28(3.43)	18.67(1.2)	22.37(4.30)	19.8(3.63)
Mean body mass (SD) (kg)	64.8(21.5)	85.0(11.3)	55.0(15.7)	49.5(4.1)
Mean height (SD) (m)	1.61(14.2)	1.79(1.9)	1.50(5.3)	1.53(7.1)
BMI (kg/m^2)	23.3(4.0)	26.4(4.3)	23.7(5.0)	21.0(2.1)

SD; standard deviation, yrs; years, kg; kilogrammes, m; metres, BMI; body mass index GMFCS; Gross Motor Function Classification System

Nine participants were tested at T0. One participant in GMFCS I did not return (contact lost) for T1; this participant was not considered in the analysis. With regards to the habitual physical activity measured by the activity monitor (ActivPAL^{TM}), only six participants had their data for both baseline and session 2. Two of the participants' physical activity data were missing due to the failure of their activity monitors to record their activity.

The raw data (mean and standard deviations) for the OMs used in this study are shown in Table 5.2. The reliability (ICC values) results based on the raw data (Table 5.2) are presented in Table 5.3. Good reliability based on the criteria suggested by Portney (2000) were observed for TUG (ICC=0.95) (95% CI 0.77–0.99), 10m SRT (0.92) (95% CI 0.44–0.97), hip extensor strength, habitual physical activity for sitting/lying, standing, stepping (0.76–0.86) (95% CI 0.02–0.98) with the exception of the sit-to-stand transitions derived from the activity monitor (ActivPAL^{TM}) (ICC=0.75) (95% CI -0.03–0.96) which indicated moderate reliability. For the other OMs, i.e. hip abductor and knee extensor strength, and PCS (derived from the SF-12), the reliability was moderate, with ICCs ranging from 0.54 to 0.74 (95% CI -0.28–0.96). Only MCS and RSES were found to have poor reliability, with ICCs of 0.45 (95% CI -0.45–0.90) and 0.13 (95% CI -0.69–0.81), respectively (Table 5.3).

Agreement as indicated by SEM for the TUG and 10m SRT were 2.48 secs and 1.3 (shuttles) respectively. For ISM, the smallest SEM was found for hip abductor strength at 0.16Nm/kg, with the highest found for knee extensor, at 0.27Nm/kg. The SEM for the step counts was 962 steps/day (where the mean score was 8695 steps/day). From the SEM, MDC_{95} was derived, which is considered to be the minimal amount of change that is not likely (95% certainty) to be due to chance variation in the measurement. The MDC thus provides information on the minimal difference needed to consider the change as being a ‘true’ change. Values for the MDC_{95} in the units of measurement and MDC_{95}% (dimensionless) are given in Table 5.2. The highest MDC_{95}% found in the current study was for knee extensor strength, with 80.58%, and the lowest was RSES, with only 8.46% (Table 5.2).

Table 5.2 Mean (SD) of the outcome measures for healthy controls and participants with CP at baseline (T0) and second assessment (T1)

		Healthy	Cerebral Palsy
TUG (seconds)	T0	6.7(0.9)	18.6(12.6)
	T1	6.6(0.8)	15.8(9.9)
10m SRT (shuttles)	T0	15.0(4.0)	8.6(4.0)
	T1	15.0(4.0)	9.0(3.0)
HE (Nm/kg)	T0	2.06(0.88)	1.32(0.55)
	T1	2.16(0.76)	1.52(0.70)
HA (Nm/kg)	T0	1.05(0.24)	0.57(0.31)
	T1	0.97(0.26)	0.58(0.29)
KE (Nm/kg)	T0	1.43(0.37)	0.85(0.36)
	T1	1.36(0.23)	1.02(0.51)
Sitting/lying*	T0	19.0(1.6)	21.6(1.4)
(hours/day)	T1	18.8(1.6)	21.0(1.6)
Standing	T0	3.3(1.2)	1.8(1.0)
(hours/day)*	T1	3.7(0.9)	2.1(1.2)
Stepping	T0	1.6(0.5)	0.7(0.4)
(hours/day)*	T1	1.8(0.7)	0.8(0.4)
Step counts*	T0	8236(2635)	2967(2391)
(steps/day)	T1	9874(4136)	2967(2391)
Sit to stand*	T0	45(10	33(10)
(number/day)	T1	45(10)	34(10)
PCS	T0	56.6(3.5)	42.4(8.0)
	T1	55.7(3.7)	40.1(10.3)
MCS	T0	45.5(7.8)	49.7(10.7)
	T1	50.1(6.8)	55.7(7.6)
RSES	T0	23(4.0)	23(4.0)
	T1	23(5.0)	26(1.0)

TUG, Timed up and go test; 10m SRT, 10m Shuttle run test; HE, hip extensor strength; HA, Hip abductor strength; KE, knee extensor strength; PCS, physical component score (SF-12); MCS, mental component score (SF-12); RSES, Rosenberg Self-Esteem Scale

**data for four days*

Table 5.3 Test-retest reliability indices of consistency ($ICC_{2,1}$) and agreement (SEM and MDC)

	Healthy controls					CP				
	ICC	**95%CI**	**SEM**	**MDC_{95}**	**MDC%**	**ICC**	**95%CI**	**SEM**	**MDC_{95}**	**MDC%**
TUG (s)	0.80	0.48–0.93	0.38	1.06	15.93	0.95	0.77–0.99	2.48	6.88	37.03
10m SRT (shuttles)	0.92	0.77–0.97	1.13	3.13	21.21	0.86	0.44–0.97	1.30	3.62	42.51
Hip extension (Nm/kg)	0.36	-0.19–0.74	0.62	1.73	80.81	0.89	0.56–0.98	0.22	0.60	45.87
Hip abduction (Nm/kg)	0.57	0.08–0.84	0.16	0.45	44.58	0.65	-0.28–0.92	0.16	0.44	76.69
Knee extension (Nm/kg)	0.16	-0.39–0.62	0.28	0.78	56.42	0.58	-0.14–0.90	0.27	0.74	80.58
Sitting/lying (hours/day)*	0.50	-0.11–0.84	1.10	3.06	16.18	0.80	0.15–0.97	0.65	1.80	8.46
Standing (hours/day)*	0.38	-0.25–0.78	0.83	2.29	68.59	0.76	0.02–0.96	0.54	1.49	77.05
Stepping (hours/day)*	0.39	-0.24–0.79	0.50	1.37	81.18	0.86	0.3–0.98	0.15	0.42	56.61
Step counts (steps/day)*	0.24	-0.39–0.72	3037	8418	92.96	0.81	0.14–0.97	962	2667	75.79
Sit to stand (numbers/day)*	0.46	-0.16–0.82	6.69	18.54	43.09	0.75	-0.03–0.96	4.89	13.56	40.32
PCSᵘ	0.53	0.03–0.82	2.43	6.73	11.98	0.74	-0.03–0.96	4.76	13.21	32.77
MCSᵘ	0.57	0.08–0.84	4.98	13.80	28.89	0.45	-0.45–0.90	8.03	22.26	41.26
RSES	0.96	0.88–0.99	0.91	2.52	11.01	0.13	-0.69–0.81	2.73	7.56	30.69

*ICC, Intraclass Correlation Coefficient; CI, Confidence Interval; SEM, Standard Error Measurement; MDC, Minimal Detectable Change; TUG, Timed Up and Go test; 10m SRT, 10-metre shuttle run test; GMFM, Gross Motor Function Measure; GPS, Gait Profile Score; COPM, Canadian Occupational Performance Measure; FAQ, Gillette Functional Assessment Questionnaire; PCS, Physical Component Score; MCS, Mental Component Score; RSES, Rosenberg Self-Esteem Scale, *derived from activity monitor known as ActivPAL™for four days, µ derived from Short Form 12 version 2 (SF-12)*

5.4.2 The test-retest reliability of physical function OMs (TUG, SRT, ISM), habitual physical activity (activity monitor), quality of life (SF-12) and self-esteem (RSES) in age-matched healthy controls

Eighteen (11 female) participants were recruited for the healthy control group; age was 20.5±4.0 years. Eighteen participants were assessed at T0. Four participants failed to return for T1 due to clinical placement (n=2) and school examination (n=2). Of the 14 participants who came for both sessions, three sets of ActivPAL^{TM} data were missing at session 2 (T1) due to a failure of the activity monitor (ActivPAL^{TM}) to capture the activity (n=2) and the participant being unable to wear it due to an examination commitment (n=1). Hence, test-retest reliability data were only available for 14 participants, with the exception of data for habitual physical activity, for which 11 sets of participant data were analysed. The demographic data of the healthy participants is shown in Table 5.4.

Table 5.4 Demographic data of age-matched healthy controls (n=14)

	Overall	Male n=7	Female n=11
Mean age (years)	20.5(4.0)	17.4(1.17)	24.3(0.44)
Mean body mass (kg)	68.10(14.6)	76.20(18.0)	63.40(10.99)
Mean height (m)	1.71(8.5)	1.76(6.2)	1.59(7.8)

Kg; kilogrammes, m; metres

With regard to the healthy control group, based on the ICC values, good reliability was shown for TUG, 10m SRT and RSES, with ICCs of 0.79, 0.92 and 0.96, respectively (Table 5.3). Moderate reliability was found for sedentary behaviour and SF-12 (both PCS and MCS). Poor reliability with an ICC <0.50 was found for all isometric strength tests, standing, stepping, step counts and sit-to-stand transitions. The SEM value for the TUG was 0.38 secs (where the mean value was 6.64 secs) and for the 10m SRT it was 1.13 (shuttles). The highest MDC% found in the current study was 94% for step count; in contrast, the lowest MDC% was 11.01% for RSES.

5.5 Discussion

The focus of this study was to investigate the test-retest reliability of the OMs for physical function (TUG, ISM, 10m SRT), habitual physical activity, quality of life and self-esteem in young people with CP and age-matched healthy controls. The values for reliability and measurement error were all derived using a test-retest study design with an interval period of approximately 6 weeks between tests. This is the first study to determine reliability indices for the physical function, quality of life and self-esteem OMs in young people with CP and age-matched healthy controls aged between 16 and 25 years.

Because of the small sample sizes of the groups, difference between the average body mass cannot always be avoided. The CP subgroups were very small, with only two males with a higher body mass in the GMFCS level I group compared to the three females with GMFCS level III. The large differences between the values for average body mass imply that the group characteristics differed between the groups, which could have confounded the outcomes of the subgroup analysis.

Overall, the ICC values suggest the majority of the PA OMs were more consistent in the CP groups than for the healthy controls. The reason for this finding may be due to the fact that healthy controls have the option of being more physically active, but they can also undertake less physical activity and be more sedentary if they so wish. Many people with CP may not have such a range of physical activity/behaviour as compared to that seen in their healthy peers.

Both healthy controls and the CP group showed good test-retest reliability for the TUG test, with ICCs of 0.8 (95% CI 0.48–0.93) and 0.95 (95% CI 0.77–0.99), respectively. Our reliability results confirmed the previous results reported for this OM in the CP population (test-retest reliability) with a one-week interval: Dhote et al. (2012) reported an ICC of 0.99 (95% CI was not reported), Williams et al. (2005) reported an ICC of 0.98 (95% CI 0.91–0.99) and Gan et al. (2008) reported an ICC of 0.99 (0.98–0.99).

The MDC value for the TUG test was calculated to be 6.88 secs for the participants with CP in this study. This value is slightly lower than those recorded in a recent study by (Carey et al. 2016) in children with CP. They reported an MDC_{95} value of 8.74s in children aged 3–10 years with CP with GMFCS I–III. A study of older children with Acquired Brain Injury (mean age of 11 years, 11 months) classified as GMFCS I–II revealed considerably lower MDC_{95} (1.2 secs), and this may be due to the shorter intervals of between 24 and 36 hours (Baque et al. 2016) in comparison to those in the current study.

The reliability for the 10m SRT tested in the CP group was also good in the current study, with an ICC of 0.86 (95% CI 0.44–0.97), which is consistent with that found in previous work by Verschuren et al. (2006), who reported ICCs of 0.97 and 0.94 (95% CI was not reported) for GMFCS I and II, respectively, and an ICC of 0.98 (95% CI 0.93–0.99) for GMFCS III (Verschuren et al. 2011). The current study acknowledges that, ideally, the reliability of the 10m SRT should be analysed according to GMFCS level as the 10m SRT includes different starting points (speeds) depending on the GMFCS level, i.e. GMFCS I starts at 5km/h, and GMFCS II starts at 2.5km/h. However, due to the restriction imposed by small sample size in the current study, this was not possible. Future studies with a higher number of participants allowing for investigation of the reliability of the 10m SRT for different GMFCS levels are therefore warranted.

The MDC value for the 10m SRT was calculated as 3.6 shuttles, and therefore a difference of at least 4 levels is required to be sure that the difference in the fitness level in a participant with CP is beyond measurement error. To our knowledge, this is the first study to explore the measurement error of the 10m SRT in young people with CP. Thus, this study provides further evidence on the reliability (consistency) of the 10m SRT.

The current study found good to moderate reliability in the CP group, with ICCs of 0.89 (95% CI 0.56–0.98) for hip extensor, 0.65 for hip abductor (95% CI -0.28–0.92) and 0.58 (95% CI -0.14–0.90) for knee extensor. The results of good reliability for the hip extensor in the current study were consistent with those found in previous work by Seniorou (2002), who reported an ICC of 0.79 (95% CI not reported) for hip extensor strength. However, contrary to the current study, they also reported good reliability for hip abductors and knee extensors, with ICCs of 0.79 and 0.84, respectively. Seniorou (2002) included the participants' age, ranging from 5 to 6 years, with a one-week interval using handheld myometry. The differences in the ICCs between this study and the previous study could be due to a longer time interval in our study, differences in measurement technique and the range of strength values.

The MDCs for isometric strength of hip extensor, hip abductor and knee extensor in the current study ranged from 0.77 to 0.44 Nm/kg. The MDC of lower limb strength in the CP population has been reported in previous studies; however, comparison with the current study is difficult as the previous studies reported non-normalised torque values (Verschuren et al. 2008b) and one study reported N/kg (Willemse et al. 2013). However, the MDC% ranging from 50% to 84% is comparable with the study by Verschuren et al. (2008) with a similar population (participants with CP, mean age of 11.1 years, GMFCS I–II) with MDC% calculated as 72%–77%.

The reliability of the habitual physical activity captured by the same activity monitor (i.e. ActivPAL^{TM}) was found to be good in CP. Our ICC finding of 0.81 (95% CI 0.14–0.97) for step counts was in agreement with a study by Bania (2014) of participants with CP and a mean age of 18.6 years with GMFCS II–III. However, our ICCs were higher for sedentary behaviour (0.80, 95% CI 0.15–0.97) and standing (0.76, 95% CI 0.02–0.96) than those reported in the previous study (Bania 2014), with moderate ICCs of 0.66

(95% CI -0.81–0.19) for sedentary behaviour and 0.60 (95% CI -0.21–0.95) for standing.

A change in the number of hours per day of at least 2.14 (MDC) on the time spent sitting or lying, as measured by the ActivPAL^{TM}, would be required in order to see a real difference in the sedentary behaviour of the CP participants in the present study. This value is similar to that observed in a repeatability study with a similar population conducted by Bania (2014). In that study, the MDC was calculated to be 2.55 hours/day (grand mean 20.27±1.58, ICC 0.66, SEM 0.92). In terms of stepping, the current study revealed a somewhat higher MDC (3049 steps/day) in comparison with that in Bania (2014), who reported MDC as 2624 steps/day (grand mean 4383.5±26262 steps/day, ICC 0.87, SEM 946.82 steps/day). In contrast, we found a slightly lower MDC for the time spent standing (MDC 1.68 hours/day in our sample compared to Bania's study, which established an MDC value of 2.2 hours/day (grand mean 2.77 hours/day, ICC 0.60, SEM 0.79 hours/day)).

The test-retest reliability for PCS was good (ICC 0.74, 95% CI -0.03–0.96), whereas it was poor for MCS (ICC 0.45, 95% CI -0.45–0.90). Although test-retest reliability has been demonstrated for PCS and MCS in older populations (Ware et al. 1996), the current study is the first to demonstrate test-retest reliability for the SF-12 among young people with CP. The ICC of PCS in the CP group of the present study is consistent with studies among patients with stroke (Bohannon et al. 2004) and rheumatoid arthritis (Hurst et al. 1998). Bohannon (2004) reported an ICC of 0.80 (95% CI was not reported) for PCS in a study of 31 stroke survivors with a mean age of 66.5 years and a mean interval time of 16.2 days. Another study involving 233 participants with rheumatoid arthritis aged between 21 and 87 years reported ICCs of 0.75 (95% CI 0.64–0.87) and 0.71 (95% CI 0.60–0.83) for PCS and MCS, respectively, with an interval of over 3 months (Hurst et al. 1998).

For the PCS and MCS, the MDCs were calculated to be 12.56 and 18.58, respectively, and therefore a difference of 13, 19 or more points, respectively, would be required to see a real difference in the quality of life of a participant with CP. The MDCs calculated in the previous study by Hurst (1998) were 13.44 (PCS) and 18.36 (MCS) in the rheumatoid arthritis population, which are similar to those found in the current study.

The current study reported poor reliability (ICC 0.11, 95% CI -0.69–0.81) of RSES in the group of participants with CP. With an MDC value of 6.29 for the RSES, this means that a difference of at least 7 points is required to detect a real change in the self-esteem score of young people with CP. So far, no studies have presented the MDC value of RSES in the CP population. As this is the first measurement of error values presented for RSES in young people with CP, it will perhaps provide reference values for future studies, although the current study does have the limitation of a low sample size.

5.6 Limitations

The healthy controls were recruited from a convenience sample and therefore may not be representative of the general population, although they were representative of young people (adolescents/young adults) attending high school and university in Musselburgh. The main limitation of the study with regards to the participants with CP and controls was the low number of participants, which resulted in a ‘poor’ rating for methodological quality in the original COSMIN rating (Mokkink et al. 2011). With regard to the original COSMIN rating, a sample size >100 rates as excellent, 50–99 as good, 30–49 as fair and <30 as poor. These ratings by COSMIN, however, were initially developed for questionnaires and therefore the sample size requirement using these guidelines may be questionable for different types of OMs. Further, neuropaediatric field studies are often limited by small sample size (Ammann-Reiffer et al. 2014, Saether et al. 2013), However, the large number of participants (>100) required to achieve a COSMIN rating of excellent might be achievable if the potential participants were identified from

multiple rehabilitation centres/clinics across the region. In terms of habitual physical activity data, only data for four of the participants were available for analysis, and therefore the results need to be treated with caution. Another possible limitation is that in the current study, the parents were permitted to complete the questionnaires (SF-12 and RSES) on behalf of the participant (if needed), and this may have affected the QoL and self-esteem scores. With regard to COPM, the participants were not blind to their previous scores; as such, bias or overestimated improvement may have occurred.

5.7 Conclusion

The TUG, 10m SRT, isometric hip extensor strength and habitual physical activity (sedentary behaviour, standing, stepping, step counts, sit to stand) derived from the activity monitor (ActivPAL^{TM}) were found to have good reliability in young people with CP. In the current study, we found that only TUG, 10m SRT and RSES were shown to have good test-retest reliability in healthy controls. The MDC values reported in this study may be of use to clinicians in helping to interpret any change in their results as being either a real change or due to measurement error in young people with CP with regard to their physical function and habitual physical activity derived from activity monitor (ActivPAL^{TM}) measures. The same does not apply, however, for QoL and self-esteem, as more studies are needed to investigate these two measures.

CHAPTER 6 PHYSICAL FUNCTION, HABITUAL PHYSICAL ACTIVITY, QUALITY OF LIFE AND SELF-ESTEEM IN YOUNG PEOPLE WITH CEREBRAL PALSY AND AGE-MATCHED CONTROLS: A COHORT STUDY

6.1 Introduction

It is well established that children with CP have decreased muscle strength (Brown et al. 1991, Elder et al. 2003, Engsberg et al. 2000), show decreased physical activity levels (Bjornson et al. 2007, Pirpiris and Graham 2004, Van den Berg-Emons et al. 1995) and report a lower quality of life (Bjornson et al. 2008) compared to typically developing children. The majority of the studies above included children aged below 16 years; however, little is known regarding physical function, habitual physical activity, quality of life (QoL) and self-esteem in young people with CP just before and after the transition to adult health services (16–25 years of age) in comparison with their age-matched peers.

6.2 Study aims

- To compare the physical function, habitual physical activity, QoL and self-esteem between young people with CP (16–25 years of age) and age-matched healthy controls.

6.3 Methods

For the CP group, the participants were identified from the patient's database of the Anderson Gait Analysis Laboratory, SMART centre, Astley Ainslie, Edinburgh. The inclusion criteria for the CP group were (a) individuals with CP between 16 and 25 years of age, (b) able to ambulate 100 metres with or without aids. The participant or carer (on behalf of the participant) were

asked to fill in the questionnaires (SF-12 and RSES) before the baseline assessment took place. Patients were excluded from the study if they had any of the following: (a) insufficient cognitive ability to give informed consent, understand and provide answers to the questions in the questionnaire booklet, (b) medical contraindications preventing them from participating in exercise testing (American College of Sports Medicine 1991), and (c) orthopaedic surgery or Botox injection in the last 6 months. The participants with CP for this cohort study comprised those in both the control and intervention groups of the RCT (into the effects of a community exercise programme) (Study 3, Chapter 7).

For the age-matched healthy controls, a convenient sample of 18 participants was recruited from Queen Margaret University (QMU) through online advertisement in the QMU moderator and from among secondary school pupils through the manager of local rugby clubs near Musselburgh and Edinburgh, with ages ranging between 16 and 25 years. Participants were excluded if they had any neuromuscular, musculoskeletal or cardiopulmonary disease which could affect their physical function. This study was approved by the QMU Research Ethics Board and National Health Services (NHS) research ethics committee (see Appendix 8).

The OMs used for this cohort study are physical function (TUG, 10m SRT, isometric strength test), habitual physical activity (ActivPAL^{TM}), quality of life (SF-12) and self-esteem (Rosenberg Self-Esteem Scale).

6.3.1 Statistical analysis

Statistical analysis was performed using Statistical Package for Social Science (SPSS) version 21. The data were retrieved from the baseline assessment dataset of 1) Study 3 for the CP group, and 2) Study 2 for the healthy controls. Due to the small sample size, comparisons of differences between groups were analysed using non-parametric tests. Firstly, the Mann-Whitney test was performed to compare the mean differences for measures; TUG, 10m SRT, isometric muscle strength, habitual physical activity

(sitting/lying, standing, step counts, stepping, sit to stand), QoL and self-esteem between the healthy and CP groups. The level of statistical significance was set at $p<0.05$. Secondly, to establish the mean differences of all the measures based on functional mobility, a Kruskal-Wallis test was performed. For this analysis, comparison of the three groups – (1) healthy group, (2) participants with CP rated level I on the GMFCS, and (3) participants with CP rated at levels II and III on the GMFCS – were performed. Participants with CP classified as GMFCS levels II and III were combined for the analysis between groups due to the small sample size of these groups. If significant differences were found between groups, a post-hoc test (Mann-Whitney) was performed to determine individual group differences with a Bonferroni adjustment. This resulted in an alpha level of 0.016.

For the habitual physical activity data derived from the ActivPAL^{TM}, participants' data were included for analysis if at least four full days (24 hours each) of data were available. The data were visually inspected and from this it was subjectively judged whether or not the ActivPAL^{TM} was to be worn. The data for time spent sitting/lying was reported over a 24-hour period and thus is inclusive of nocturnal sleeping time and non-wearing time. The current study also did not consider weekdays and weekend days when analysing the data. However, the majority of the participants wore the monitor for seven days, hence including both weekdays and weekend days.

6.5 Results

Seventeen participants with CP and 18 age-matched healthy controls were tested at baseline. One healthy participant's activity monitor (ActivPAL^{TM}) failed to capture the physical activity data; therefore, only 17 sets of physical activity data were analysed for the healthy control group. The demographic data of the participants with CP and healthy controls are presented in Table 6.1.

As expected, the healthy participants achieved scores indicating a higher level of physical function than the CP group (all GMFCS levels), with statistically significant differences between the two groups found for the TUG ($p=<0.001$), 10m SRT ($p=0.01$), hip extensor strength ($p=0.03$), hip abductor strength ($p<0.001$), knee extensor strength ($p<0.001$) and all habitual physical activity (time spent sitting/lying ($p<0.001$), time spent standing ($p=0.001$), time spent stepping ($p<0.001$), step count ($p<0.001$) and sit-to-stand transitions ($p=0.03$)) (Table 6.2).

Table 6.1 Demographic data of participants with CP and age-matched healthy controls

Healthy controls n=18		Participants with CP		
		Overall n=17	GMFCS I n=6	GMFCS II/III n=11
Gender (female:male)	11:7	11:6	3:3	8:3
Mean age (years)	22.73(3.6)	20.02(3.16)	19.02(2.26)	20.56(3.54)
Mean weight (kg)	69.01(14.06)	60.97(20.1)	64.5(19.46)	59.05(21.11)
Mean height (m)	1.71(8.3)	1.61(10.9)	1.68(9.9)	1.57(9.6)

CP; cerebral palsy, kg; kilogrammes, m; metres, GMFCS; Gross Motor Function Classification Score

When analysing the differences between the participants with CP classified as GMFCS level I and the healthy control group, scores indicating a statistically lower level of physical function in the CP GMFCS I group were found for TUG (p=0.01), 10m SRT (p=0.01), hip abductor strength (p=0.002) and knee extensor strength (p=0.005) (Table 6.2). Comparison among GMFCS levels showed the time required to complete the TUG significantly differed between GMFCS levels I and II/III (p=0.012). Time increased with increasing severity. In addition, hip extensor strength for the participants in the CP group with GMFCS level I was significantly higher than in the CP group with GMFCS levels II/III (p=0.015). No significant differences were found between GMFCS level I and levels II/III in other physical function measures, i.e. 10m SRT (p=0.25), hip abductor strength (p=0.13) and knee extensor strength (p=0.55), as well as habitual physical activity (time spent sitting/lying (p=0.07), time spent standing (p=0.48), time spent stepping (p=0.23), step counts (p=0.19) and sit-to-stand transitions (p=0.056).

With regards to QoL, the PCS derived from the SF-12 was significantly lower in the CP group in comparison with healthy peers (p=0.043) (refer to Table 6.2). The mean score of MCS in the CP group was lower than for the healthy controls (45.72 versus 48.44); however, this was not statistically significant (p=0.25) (Table 6.2). Investigation between GMFCS level I and levels II/III also showed a significant difference in PCS scores, showing that the participants with GMFCS levels II/III reported lower scores in the physical component of QoL compared to those with GMFCS level I (p=0.016). With regards to self-esteem, no significant difference was found between the CP and healthy control groups, neither was there any significant difference between the GMFCS levels.

Table 6.2 Physical function, habitual physical activity, quality of life and self-esteem in the group of young people with CP and age-matched healthy controls

			Mean (SD)			
Outcome measures			**Healthy controls[μ]**	**CP group**		
				CP overall n=17	**GMFCS I n=6**	**GMFCS II/III n=11**
Physical function	TUG (s)		6.62(0.85)	18.17(11.81)[a]	9.33(2.74)[bc]	22.99(12.13)[d]
	10m SRT (shuttles)		15.0(3)	9.0(5.0)[a]	8.0(6.0)[bc]	9.0(4.0)
	Isometric strength tests	Hip extension (Nm/kg)	2.03(0.82)	1.17(0.49)[a]	1.60(0.34)[c]	0.98(0.42)[d]
		Hip abduction (Nm/kg)	1.22(0.44)	0.57(0.29)[a]	0.68(0.23)[bc]	0.50(0.31)
		Knee extension (Nm/kg)	1.47(0.35)	0.84(0.30)[a]	0.92(0.32)[bc]	0.80(0.30)
Physical activity	Habitual physical activity	Sitting/lying (hours/day)	18.88(1.39)	20.98(1.54)[a]	20.08(1.89)[c]	21.47(1.12)
		Standing (hours/day)	3.30(1.20)	1.97(0.82)[a]	2.24(0.75)[c]	1.82(0.86)
		Stepping (hours/day)	1.58(0.50)	0.72(0.40)[a]	0.91(0.46)[bc]	0.61(0.86)
		Step counts (steps/day)	8575(2763)	3421(2396)[a]	4628(2926)[bc]	2762(1886)
		Sit-to-stand transitions	42.94(9.94)	36.06(12.50)[a]	44.84(14.37)[c]	31.27(8.68)
Quality of life	SF-12	PCS (34.75–56.71)	56.14(3.54)	42.11(8.87)[a]	48.17(7.11)[c]	38.82(8.2)[d]
		MCS (15.37–62.39)	47.78(7.59)	45.72(13.57)	40.86(17.78)	48.38(10.71)
Self-esteem	RSES (0-30)		22.89(4.32)	19.76(6.69)	17.50(9.35)	21.00(4.80)

[μ]n=18 for all OMs in exception for physical activity n=17
[a]Healthy versus CP (all GMFCS levels) p<0.05; [b]Healthy versus GMFCS level I p<0.016; [c]Healthy versus GMFCS levels II/III p<0.016; [d]GMFCS level I versus GMFCS levels II/III p<0.016

6.6 Discussion

The aim of this study was to compare the physical function (TUG, ISM, 10m SRT), habitual physical activity, QoL and self-esteem in young people with CP and age-matched healthy controls. The present study shows that the physical function (TUG, 10m SRT, isometric muscle strength), habitual physical activity and the physical component of QoL between young people with CP and age-matched healthy controls were significantly different. The TUG test results, hip extensor strength and PCS were also significantly different between the two groups with different GMFCS levels (I<II/III, $p<0.05$).

As expected, the healthy participants showed faster TUG times in comparison with the participants with CP in this study. The CP group took an average of 18.17 secs to complete the TUG test, which was nearly three times the average time taken by the healthy group (6.62 secs, $p<0.05$). Zaino (2004) also compared the TUG test between healthy controls and children with CP (mean age 10.9±0.7) classified as GMFCS I–III and reported similar findings. The average time taken for the healthy controls was 5.1 secs in the study by Zaino (2004), thus similar to the finding of the present study for the healthy controls. With regard to the CP group, Zaino (2004) reported averages of 6.1 secs and 8.1 secs in participants with GMFCS I and II/III, respectively, and these values were lower than the present study, which recorded averages of 9.3 secs and 23 secs for GMFCS level I and levels II/III, respectively. This dissimilarity could be due to the age group difference since Zaino (2004) focused on children, whereas the current study focuses on adolescents and young adults with CP. Evidence has shown than functional mobility decreases with age (Day et al. 2005).

The mean TUG score of 18.17 secs in the CP group was similar to that found in a previous study with a similar population (i.e. GMFCS I, II and III), mean age of

14.97±2.03 years with TUG scores of 18.34±9.66secs (Chrysagis et al. 2014). The results of the current study demonstrate the differences in TUG test performance between young people with CP classified as GMFCS levels I and II/III. Not surprisingly, the time to complete the test increased from GMFCS level I to level III. These findings are in agreement with previous research in which TUG scores increased between GMFCS levels I and III (Hassani et al. 2014).

The healthy control group achieved a statistically significant higher number of shuttles in the 10m SRT compared to the number achieved by the CP group ($p=0.001$). The healthy controls reached an average of 15 shuttles in the 10m SRT. The CP group achieved averages of 8 and 9 shuttles for GMFCS I and II/III, respectively. The CP group with GMFCS level I and healthy controls started at 5km/h, while the CP group with GMFCS II/III started at 2.5 km/h. As such, comparison between the different GMFCS levels is difficult. The reference values (number of shuttles) in relation to height for the 10m SRT have been reported by Verschuren et al. (2010) based on their study of participants with CP ranging from 6 to 19 years of age (mean 12 years) with GMFCS I and II. In the current study, the average height of the participants with CP classified as GMFCS I was 179.5 cm, and the average number of shuttles was 8, which means that the participants' achievements fell between the 25^{th} and 50^{th} centiles for their height (Verschuren et al. 2010). Regarding the participants with GMFCS II in the current study, the 10m SRT revealed their fitness level to be around the 50^{th} centile (i.e. average). The usage of this reference value is recommended to keep track of fitness levels in individuals with CP with GMFCS I and II.

With regard to isometric muscle strength, in the current study, the CP group showed statistically significant lower values than the healthy controls, i.e. hip extensor (1.17 vs 2.3 Nm/kg), hip abductor (0.57 vs 1.22 Nm/kg) and knee extensor (0.84 vs 1.47 Nm/kg). Our findings of significant lower strength values in the CP group compared to healthy controls are in agreement with those

reported in previous studies (Damiano and Abel 1998, Thompson et al. 2011, Wiley and Damiano 1998). A previous study by Thompson et al. (2011) reported differences in strength between the CP group and healthy controls. In particular, the relative differences between the CP group and healthy controls in hip abduction and knee extension in the present study appear higher than those seen in Thompson et al. (2011). In the current study, we have relative differences for hip extension of 0.65Nm/kg and knee extension of 0.63Nm/kg. In comparison, Thompson et al. (2011) found differences of 0.44Nm/kg for hip extension and 0.32Nm/kg for knee extension. This is more likely due to the discrepancies in terms of GMFCS levels and age group between our study and Thompson et al. (2011). The hip abductor (0.57Nm/kg) and knee extensor (0.84Nm/kg) strength values of the CP group (all GMFCS levels) in the current study were consistent with those in previous CP studies (Thompson et al. 2011, Dallmeijer et al. 2011). Dallmeijer et al. (2011) included participants with a mean age of 18.9±2.0 years, classified as GMFCS II and III, and strength was measured using a handheld dynamometer. They reported the normalised torque for hip abductor as 0.59Nm/kg and knee extensor as 1.04Nm/kg. However, the hip extensor strength in our study (CP overall group) was higher (1.17 Nm/kg) than that in Dallmeijer et al.'s study, which reported 0.89Nm/kg. Thompson et al. (2011) also reported an almost similar torque to that found in the current study for hip abductor (0.61Nm/kg) but not for hip and knee extensor, whereby they reported higher torque (2.37Nm/kg and 1.50Nm/kg, respectively) than both the current study's finding and that of Dallmeijer et al. (2011).

Young people with CP have been reported to have lower levels of habitual physical activity compared to their healthy peers (Carlon et al. 2013). The current study reveals that the mean step count for the healthy group was 8574 steps/day, which was more than double the step count of 3420 steps/day in participants with CP ($p<0.05$). The sedentary behaviour (sitting/lying) between the healthy control and CP groups was significantly different, with the CP groups

spending 2 hours more on sedentary behaviour ($p<0.05$). Our results are in agreement with those in the study by Stevens et al. (2010), who compared the physical activity measured by the step activity monitor (Orthocare SAM^{TM}) in children with CP and typically developing (TD) children. They reported a significant difference in step counts (CP 3171 vs healthy 6812 steps/day, $p<0.001$) and time spent sitting/lying (CP 80.7% vs healthy 67.0%/day, $p<0.001$) in older children (14–18 years) in comparison with the TD group (Stevens et al. 2010). In terms of the number of hours spent on sedentary activity in the CP group, our result of 21.3 hours/day is consistent with that from a previous study by Bania et al. (2016) (participants with CP; mean age 18.9 years; GMFCS II and III). In the Bania et al. (2016) study, physical activity was recorded using the same activity monitor as that used in the current study, revealing that the average time spent sitting/lying was 20.1 hours daily, which was two hours greater than that found in the current study. Sedentary time, however, is difficult to compare between studies and even within participants, since the current study includes non-wear time.

The physical component summary (PCS) derived from the SF-12 v2 was significantly lower in the CP group compared to the healthy controls, (42.11 versus 56.14, $p=0.023$). Using the Child Health Questionnaire Child Form (CHQCH) to measure quality of life, Bjornson et al. (2008) also reported that the physical function component in the CHQCH perceived by the youth with CP was significantly lower than for the healthy controls ($p<0.01$). In terms of the mental component summary (MCS), no significant difference was found between the healthy controls and CP groups in the current study ($p=0.1$). The average PCS (42.11) and MCS (45.72) of the CP group in the present study are comparable to those in a previous study by Saebu and Sørensen (2011), which investigated the factors associated with physical activity among young adults with physical disability between the ages of 18 and 30 years. In that study, they found the

physical and mental scores to be 40.9 and 40.2 respectively (Saebu and Sørensen2011).

There was no significant difference between the healthy controls and CP groups with regard to their self-esteem scores as measured by RSES. Magill and Hurlbut 1986 examined the self-esteem of 22 adolescents with CP aged between 13 and 18 years and found no significant differences compared with healthy peers for overall self-esteem, as measured using the Tennessee Self-Concept Scale (Fitts and Roid 1964). The CP group (overall) in the present study scored an average of 21. So far, only one study has published the results of self-esteem in young people with CP, and this was a study by Manuel et al. (2003) involving participants with CP, with a mean age of 13.0 (3.0) years. Thirty-seven per cent (n=7) of the participants with CP in the current study reported a low self-esteem (scores ≤18). Lower self-esteem in young people with CP was also reported by Manuel et al. (2003), who conducted a study with pre-adolescents and adolescents with CP aged from 9 to 18 years and found similar results, with 30% scoring below the cut-off point for low self-esteem. A study investigating self-esteem from adolescence to adulthood in a healthy population indicated that self-esteem increases as individuals move out of adolescence, as this group has a less dependent relationship with their parents, a greater reliance on their peer group and a greater emphasis on education and career development (Magill-Evans and Restall 1991).

6.7 Limitations

The healthy controls were recruited from a convenience sample and therefore may not be representative of the general population, although they may be representative of those at school or university. Due to the small sample size of participants with CP, the results of the current study should be treated with caution. The current study also did not consider weekdays and weekend days

separately when analysing the data. There is a possibility of differences in the participants' levels of physical activity during weekdays in comparison to weekend days.

6.8 Conclusions

This study confirms that the young people with CP that were studied have lower physical function, habitual physical activity and physical component scores for quality of life than their age-matched peers. This study highlights important considerations with regard to future studies looking to recruit larger number of participants with CP, especially with regard to those before and after the transition to adult health care services. These results can thus serve as a benchmark for studies aiming to improve physical function, physical activity, QoL and self-esteem in young people with CP.

CHAPTER 7 EFFECTS OF AN 18-WEEK PRAGMATIC COMMUNITY EXERCISE PROGRAMME ON THE PHYSICAL FUNCTION, GAIT QUALITY, HABITUAL PHYSICAL ACTIVITY, QUALITY OF LIFE AND SELF-ESTEEM OF YOUNG PEOPLE WITH CP: A SINGLE-BLIND RANDOMISED CONTROLLED TRIAL (RCT)

7.1 Introduction

In Chapter 2, the evidence regarding the efficacy of exercise interventions for young people with CP was explored through a narrative review, and gaps were identified. In addition, Chapter 2 described potential outcome measures. Several potential outcome measures which may be used to assess the effects of an exercise programme were explored in terms of their reliability, validity and feasibility. This study aims to address some of the gaps in this evidence by evaluating the effects of an 18-week pragmatic community exercise programme on the physical function, gait quality, habitual physical activity, quality of life and self-esteem of young people with CP. The primary end point for this study was selected as 6 weeks as this interval has been suggested as the time over which the effects of an exercise programme on physical fitness occur (Taylor et al. 2013). In addition, this primary end point will maximise the possible sample size for analysis as dropout is common in exercise studies.

7.2 Research questions

1) What are the effects of a 6-week pragmatic community exercise programme on the outcomes of physical function (Timed Up and Go Test, 10m Shuttle

Run Test, isometric muscle strength, Gross Motor Function Measure 66) and gait quality (Gait Profile Score) compared to a usual care control group?

2) What are the effects of a 12-week pragmatic community exercise programme on the outcomes of objective habitual physical activity patterns, self-reported function (Canadian Occupational Performance Measure, Gillette Functional Assessment Questionnaire), quality of life and self-esteem compared to a usual care control group?

3) How does exposure to a pragmatic community exercise programme affect the outcomes of physical function and gait quality at 6, 12 and 18 weeks compared to the outcomes at baseline?

4) How does exposure to a pragmatic community exercise programme affect the outcomes of objective habitual physical activity patterns, self-reported function, quality of life and self-esteem at 12 weeks compared to the outcomes at baseline?

5) What is the feasibility of an 18-week pragmatic community exercise programme in adolescents and young adults with CP?

6) What is the perception of adolescents and young adults with CP with regard to an 18-week pragmatic community exercise programme?

7.3 Null hypotheses

H_0 1 – Exposure to 6 weeks of a pragmatic community exercise programme will not elicit statistically significant differences in the outcomes of physical function and gait quality compared to a usual-care control group.

$H_0$2 – Exposure to 12 weeks of a pragmatic community exercise programme will not elicit statistically significant differences in the outcomes of objective habitual physical activity patterns, self-reported function, quality of life and self-esteem compared to a usual-care control group.

$H_0$3 – Exposure to 18 weeks of a pragmatic community exercise programme will not elicit statistically significant differences in the outcomes of physical function and gait quality at 6, 12 and 18 weeks compared to the outcomes at baseline.

$H_0$4 – Exposure to 12 weeks of a pragmatic community exercise programme will not elicit statistically significant differences in the outcomes of objective habitual physical activity patterns, self-reported function, quality of life and self-esteem at 12 weeks compared to the outcome at baseline.

7.4 Methodology

7.4.1 Study design

The initial aim was to undertake a single-blind randomised control trial (RCT) with each participant involved over a period of 18 weeks. The study compared two groups, one receiving the exercise (intervention) and one group acting as a control group, who continued with their usual care. The participants were randomised by minimisation using GMFCS level as a factor. Randomisation was performed by the students' supervisor, who was not involved in the data collection. The researchers (CS & AZ) responsible for the data collection and analysis were blind to the group allocation.

7.4.2 Sample size calculation

The sample size calculation was based on a reported effect size of *d*=1.17 for the GMFM 66 dimensions D & E in a study investigating the effects of sit-to stand resistance exercise for children with CP (Liao et al. 2007). To achieve 80% power of detecting a difference with a significance level of 0.05, a sample size of 14 participants in each group was required. Taking into account 10% attrition, 36 participants (18 in each group) were needed for an appropriately powered RCT with one primary end point, which in this study was at 6 weeks.

7.4.3 Participants

Potentially eligible participants were identified from the patient database of the Anderson Gait Analysis Laboratory, SMART centre, Astley Ainslie Hospital, Edinburgh. The inclusion criteria were (a) individuals with CP between 16 and 25 years of age, and (b) individuals able to ambulate 100 metres with or without aids. Patients were excluded from the study if they had any of the following: (a) insufficient cognitive ability to give informed consent, understand and provide answers to the questions in the questionnaire booklet, (b) medical contraindications to participating in exercise testing (American College of Sports Medicine 1980) and (c) orthopaedic surgery (last 12 months) or Botox injection in the last 6 months. All of the participants signed an informed consent form prior to taking part and the study was approved by the QMU Research Ethics Board and National Health Services (NHS) local research ethics committee (Appendix 8). Management approval was provided by NHS Lothian Research and Development.

7.4.4 Exercise programme: Exercise logbook, Frequency of Training and Supervision

Each of the participants in the exercise group received an exercise logbook prior to the exercise programme. The exercise logbook was designed for easy

transport within the home and leisure centre. The exercise logbook included the structure of the exercise programme (Figure 7.1), information on aerobic and strength training, instructions on how to log the workouts, pictorial descriptions of the exercises, instructions on how to increase resistance and weekly exercise logs. Each participant in the exercise group was asked to log each exercise session by completing the relevant boxes in their logbook (Figure 7.1), and fitness instructors and a physiotherapist (when present) were asked to sign off each log. This allowed the researcher and physiotherapist to check the participants' fidelity to the exercise programme.

The participants in the exercise group undertook (supervised, when required, by a fitness instructor) circuit exercise training (a combination of aerobic and muscle strength exercises) at their local leisure centre or health club. On the first visit and then for between two and four further visits, a paediatric physiotherapist experienced in exercise referrals to leisure centres instructed both the participants and the fitness instructor on the content and execution of the exercises. This was done to ensure the exercises were at the appropriate level for the participants' capability and fitness. The participants were subsequently prescribed to attend the local leisure centre to exercise three times per week for the first six weeks (1–6), (ii) twice per week for weeks 7–12 (with encouragement to complete a third unsupervised session each week, and finally (iii) to attempt to complete three exercise sessions each week for weeks 13–18.

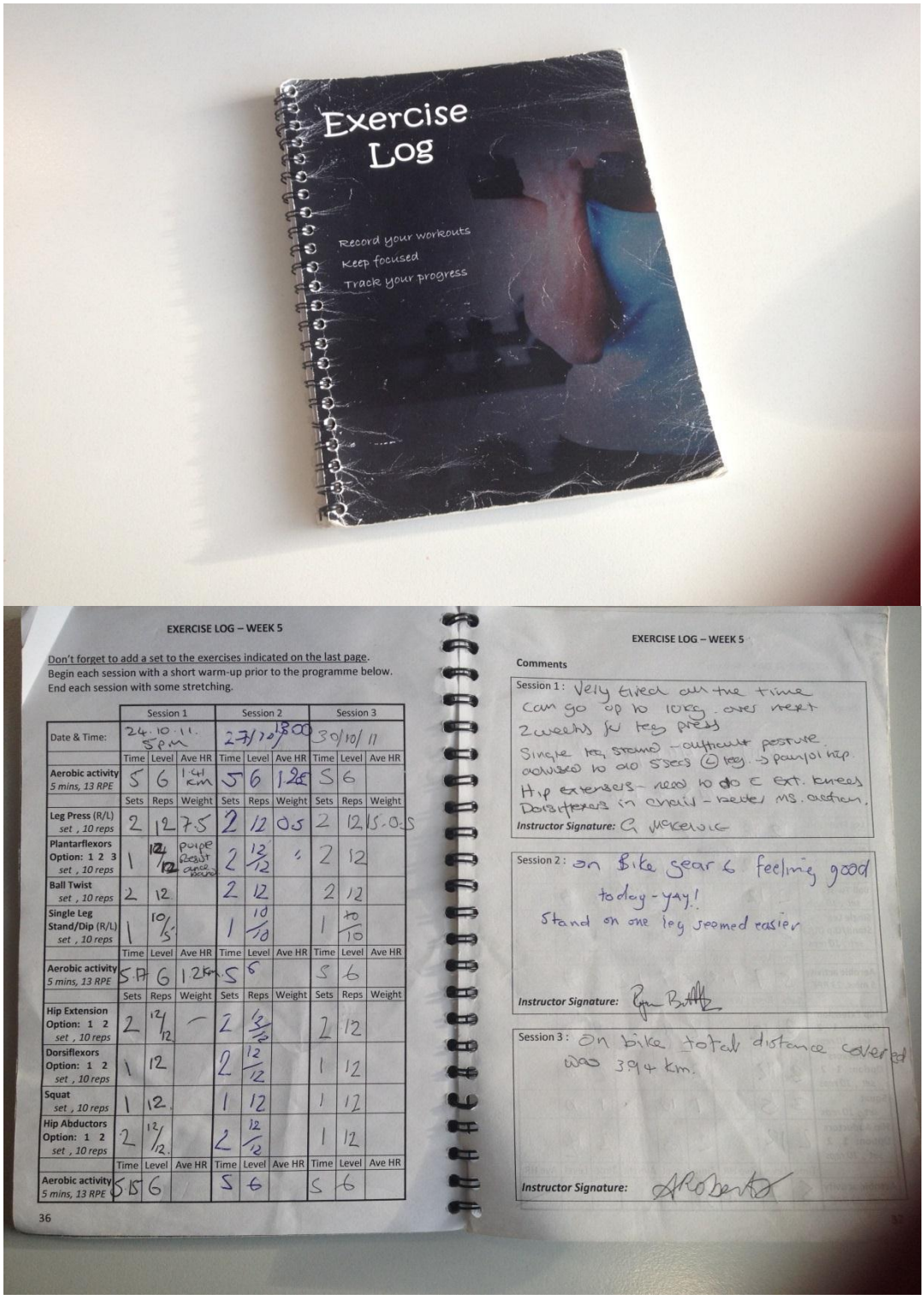


Figure 7.1 Front page of the logbook and example of a written exercise log

7.4.5 Content of the exercise programme

A schematic of the structure of the initial exercise programme is given in Figure 7.2. The choice of the particular strength exercises (i.e. how the exercises were performed) was based on the recommendation of an experienced NHS paediatric physiotherapist, and the progression of the exercise (both strength and aerobic) was based on recommendations outlined in (ACSM's exercise management for persons with Chronic Illnesses and disabilities 2003). Each visit to the leisure centre was broken into blocks of aerobic exercise interspersed with two blocks of different strength training exercises, which could be adapted to the ability of the participant (Table 7.1). At the start of the programme the participants were asked to do one set of 10 repetitions of lifting weights and this was done at the rate of 10 repetitions maximum (10RM) wherever possible. 10RM is the maximum amount of weight that can be lifted 10 times while maintaining a good form (ACSM 2003). The 10RM was performed according to the guidelines outlined by the ACSM (2003). The participants were asked to carry out a warm-up and stretching of the lower limbs prior to starting the test. Then, the participants were asked to perform the exercise for five repetitions for familiarisation. After resting for 3 to 4 minutes, the test began with the lightest weight and the participants were asked to repeat this 10 times. Once they had finished, the participants were asked whether they were able to repeat the exercise more than 10 times, and if they could, additional weights were added. The procedure was repeated up to the maximum weight that the participants could lift 10 times. A resting time of between 3 and 4 minutes was compulsory between the tests (if weight was added).

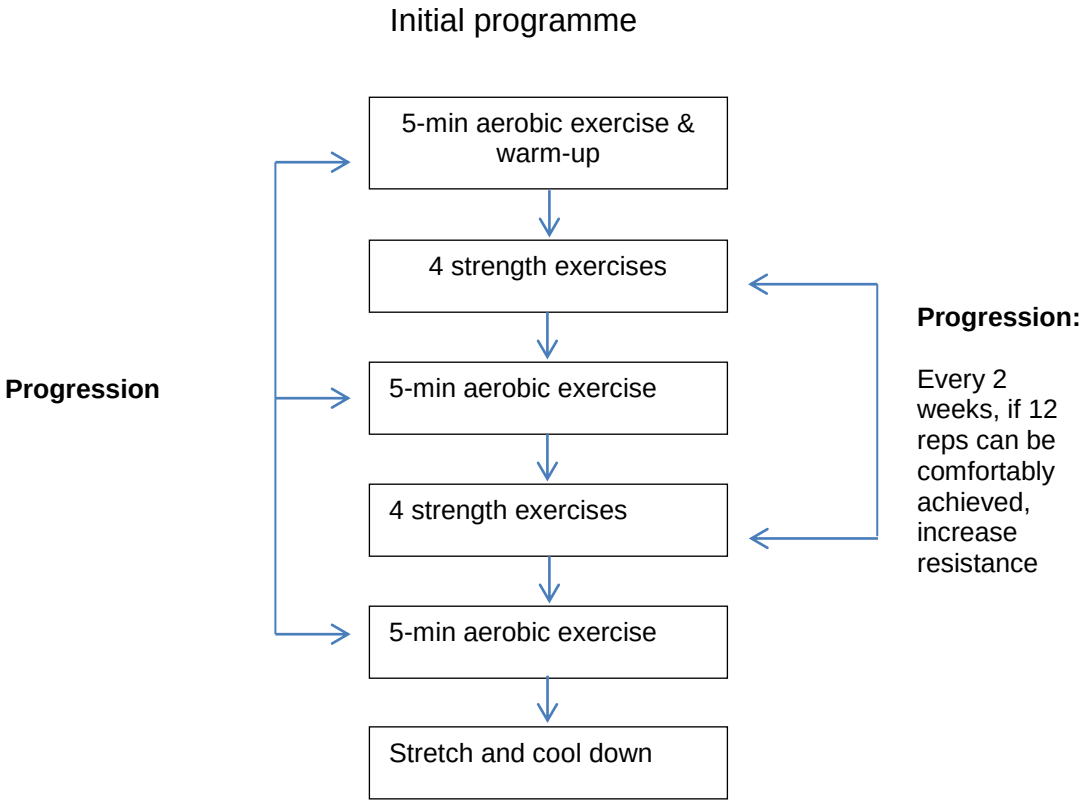
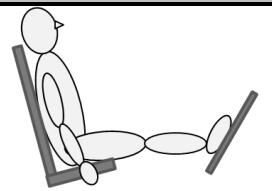
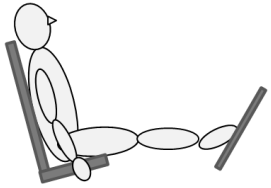
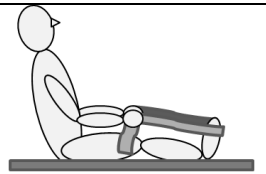
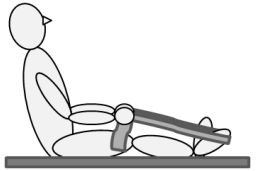
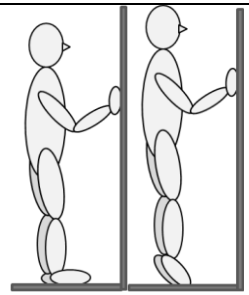
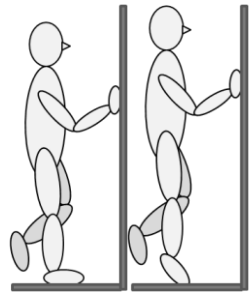


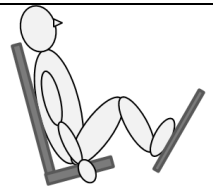
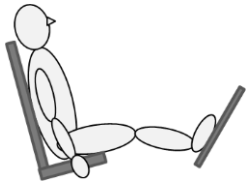
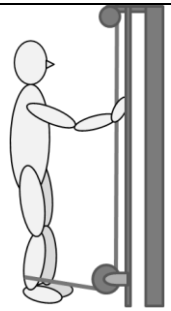
Figure 7.2 Standard exercise programme structure

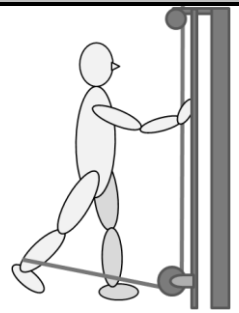
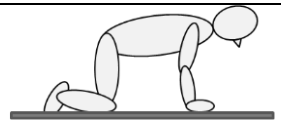
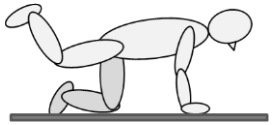
For the aerobic blocks, the participants had the option of either a reclining or upright exercise bike, treadmill, Motomed (motorised exercise bike), cross trainer or rowing machine. The participants were asked to exercise at a perceived level of exertion of 13 (‘somewhat hard’) on the 6–20 Borg Scale (Borg 1970).

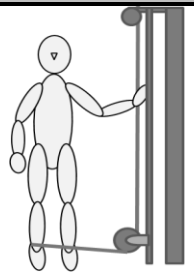
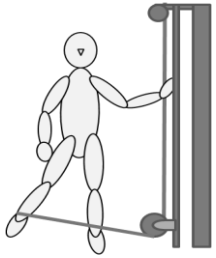
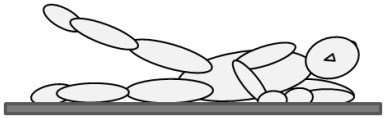
Table 7.1 Strength exercise descriptions

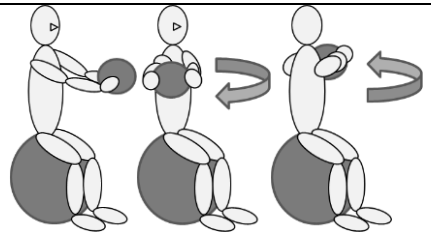
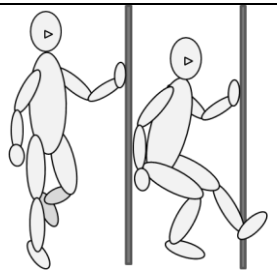
Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
Plantarflexors	Leg press machine, seat adjusted so legs are straight, press against machine with toes as if pressing a pedal	Increase resistance on the machine by 2–5% (smallest increment)	 Start Position  End position
	Alternative 1: Seated on floor, legs straight, holding TheraBand looped over foot, point toes against resistance. Each foot separately.	Shorten/double TheraBand, or use one which provides greater resistance (should be working at about 10RM)	 Start position 

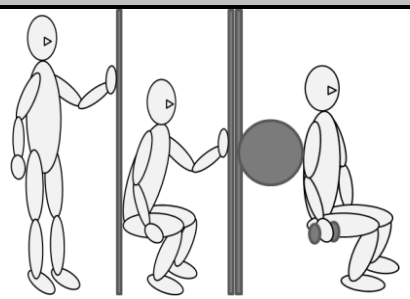
Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
			End position
	Alternative 2: heel raises, stabilise with hand(s) on wall. Each limb simultaneously or separately.	On only one leg or while holding free weight, increase resistance by 2–5%	 Start position End Position OR  Start position End Position
Dorsiflexors	Walk on heels	n/a	
	Alternative: seated on floor with legs straight, flex	n/a	

Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
	toes towards knees		
Knee extensors	Leg press machine	Increase resistance by 2–5%	 Start position  End position
Hip extensors	Standing (adjustable pulley) machine with knee raises. Each limb separately	Increase resistance by 2–5%	 Start position

Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
			 End position
	Alternative: on hands and knees, tighten glutes, lift leg. Each limb separately.	n/a	 Start position  End position

Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
Hip abductors	Standing (adjustable pulley machine) with ankle out to the side. Each limb separately.	Increase resistance by 2–5%	 Start position  End position
	Side lying, leg raises with or without TheraBand for resistance. Each limb separately.	Shorten/double TheraBand, or use one which provides greater resistance (should be working at about 10RM)	 Start position  End position

Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
	Alternative: sideways walking	n/a	
Core	Seated on ball, twist side to side, with or without weight in hands	Add/increase weight by 2–5%	 Start position Twist right Twist left
Core+lower body	Single leg standing, add dip if possible		 Start position Add dips

Muscle group	Exercise description	Progression – as required by 2-week 'self-assessment'	Illustrations
Core+lower body	Squat, with or without free weights held at sides	Add/increase weight by 2–5%	 Start position End position Variations

7.4.6 Progression within the exercise programme

At week 5, an individual component of the programme was included. The physiotherapist identified individual weaknesses to work/focus on and selected four strength exercises for which participants should complete two sets instead of only one. The structure of the programme remained the same. The participants just completed a second set of 10 reps (or as close to 10 as possible) of the chosen exercises before moving on to the next exercise.

Throughout the programme, every two weeks (including the first 4 weeks), the participants were asked to perform a 'self-assessment', which they also had the option of completing with the fitness instructor if they so wished. This assessment was used to determine whether or not to increase the resistance during an exercise. For each exercise the participants attempted to complete 12 repetitions on their last (or only) set. If 12 repetitions could be achieved comfortably, the resistance for that exercise would be increased incrementally during the following training session. For the weight machines and exercise where free weights were used as resistance, this was done by increasing the weight by the smallest increment. In cases where the TheraBand^{TM} provided resistance, the TheraBand^{TM} was shortened or doubled, or a band with greater resistance was used. If increasing the weight was not possible, then the number of repetitions was increased in order to achieve exercise progression.

Progression of the aerobic component was achieved by increasing either the duration or the level of resistance of the exercise bikes or the walking speed for the treadmill, so long as the perceived exertion remained at 'somewhat hard' (level 13).

The participants were asked to record their exercise sessions in a logbook, with the fitness instructor or physiotherapist, if present, adding his or her signature. The participants in the control group continued with their normal routine but were offered the same exercise programme after completion of the trial. Justification

of the exercise programme using the FITT principles (Heyward 2010) is shown in Table 7.2.

Table 7.2 Justification of the exercise programme using the FITT principles (Heyward 2010)

FITT principle (Heyward 2010)		Exercise programme in the present study	Justification
Frequency		3x/week for Block 1, 2x/week for Block 2	For novice/beginner it is recommended to do exercise 2–3x/week (ACSM 2003)
Intensity		10 repetitions for strength training, Borg Scale of '13' for aerobic training	The intensity and progression of the exercise are based on the guidelines from ACSM (2003)
Time	Duration of the training in each session	Approximately 1 hour	Recommended by ACSM (2003)
	Duration of the exercise programme	18 weeks	- to stimulate improvement in physical function, habitual physical activity, quality of life and self-esteem - to promote exercise behaviour
Type		Aerobic and strength training	Recommended by experienced paediatric physiotherapist and as outlined by ACSM (2003)

ACSM; American College of Sports Medicine

7.4.7 Assessments

Assessments at the QMU gait analysis laboratory took place at baseline before the exercise programme took place and at approximately 6, 12 and 18 weeks

during the programme for both groups. The six-week interval between assessments was considered an appropriate time during which intervention-induced changes in strength may be detectable in young people with CP (Taylor et al. 2004). Travel expenses to and from QMU were reimbursed. A detailed assessment protocol is given below.

Assessment Sessions

Four assessments for each participant would take place at QMU over the 18-week study. The following section describes the protocol for these sessions. The first section covers the preparation to go through before the participant arrived followed by the protocol followed during the actual data collection. Three questionnaires, namely the SF-12, RSES and FAQ, were mailed to the participants a few days before the assessment took place together with the appointment letter. In the appointment letter, the participants were instructed to fill in the questionnaires in advance and were asked to bring these with them on the day of assessment at QMU.

Preparation

The following section outlines what needed to be completed/checked prior to the arrival of the participant.

Motion analysis system set-up

- Create session and subject
- Calibrate the system and save/record camera residuals
- Reset and zero FP and ensure they are working properly
- Required equipment:
  - ✓ Markers with tape
  - ✓ Extra tape
  - ✓ Wrap/tape/Velcro strap to hold wands on
  - ✓ Clean shorts

Strength set-up

- Have straps and myometer prepared
- Required equipment:
  - ✓ One padded thigh strap with buckle
  - ✓ Two padded shin straps
  - ✓ Karabiner attached to either end of the myometer (with straps)
  - ✓ Adjustable-length strap attached to fixed rail
  - ✓ Small stool/table

Anthropomorphic measures

Required equipment:

- Measuring tape
- Lined paper, protractor, ruler, square
- Callipers

GMFM 66 (Dimensions D & E)

Ensure all required supplies are present:

- Ball
- Stick
- Ruler
- Low stool
- High bench
- Mat
- Stopwatch

Fitness testing

- Space booked (QMU sports hall)
- Floor marked with proper distance
- CD player available

ActivPAL™

- Charged and set to record
- Adhesives (Palstickies) available to hand out to participants

Data Collection

The following section describes the protocol that was followed during the data collection session once the participant had arrived.

Consent

- Ensure informed consent is obtained prior to beginning any of the testing
- Describe what will be asked of the participant and answer any questions he/she may have
- Obtain informed consent on the appropriate consent form.

Canadian Occupational Performance Measure (COPM)

Short interview with the participants to get them to identify activities of daily living that they need to do, want to do or are expected to do. Get them thinking about a number of different activities with the help of the categories on the COPM. Get the participant to rate the importance of these activities and pick out the 5 most important.

For the 5 most important measures, get the participant to rate their level of performance and satisfaction on a 10-point scale. Performance is rated from 1: 'not able to do it at all' to 10: 'able to do it extremely well', and satisfaction is rated from 1: 'not satisfied at all' to 10: 'extremely satisfied'.

To calculate a score, take the average of the performance and satisfaction scores.

Patient Information

Basic participant information should be recorded, such as:

- details of the use of any walking aids

- information on any surgery undergone
- any other conditions the participant has
- the gross motor function classification according to the guidelines for 12–18-year-olds.

Timed up and Go (TUG)

Participant is seated in a chair with arms and a backrest. Place a mark on the floor 3 m away. Ask the participant to get up, walk to the line, cross it, turn around and return to a seated position in the chair. This should be performed at a comfortable pace. Time how long this takes from when the participant begins standing to when he/she is seated again. Repeat until 3 trials are recorded.

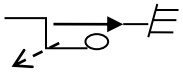
Strength Testing

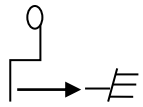
Strength is measured using a handheld myometer, fixed to a solid support. It is measured in a gravity-neutral position for each muscle group. Three trials are completed for each muscle group and the participants are given a 30s rest between trials. The myometer is zeroed with no tension on the device.

The muscle groups to be evaluated are (bilaterally):

- Hip extensors
- Hip abductors
- Knee extensors

Details for each muscle group:

Muscle group	**Position**	**Stabilisation**	**Resistance**	**Lever arm** ⊥distance from force to
Hip extensors 	Supine – Hip at 90°, knee at 90° with lower leg supported, low bench under lower leg (or held	Hip belt – attached at an angle so it pulls downward and inferiorly	Distal femur, proximal to femoral condyles	Greater Trochanter

Hip abductors 	Supine – Knee propped up on blanket to allow comfortable flexion of hip and knee and to raise	Hip belt	Distal femur, proximal to femoral condyles	Greater Trochanter
Knee extensors 	Seated, knee at 90° on chair with backrest (back of chair is pushed up against the plinth so that it is stable)	Non-tested foot on floor	Distal Shank	Lateral knee joint line

⟶ Direction of resistance from the myometer

⇢ Direction of resistance from the hip belt for stabilisation

Anthropomorphic Measures

These are basic body measures which are required for gait analysis. They include:

- Height (mm)
- Weight (Kg)
- Knee width (mm)
- Malleolar width (mm)
- Leg length (mm)
- Tibial Torsion (deg)

Knee width

Measured between the medial and lateral epicondyles using the large sliding callipers and applying gentle pressure.

Malleolar width

Measured between the medial and lateral malleoli using the large sliding callipers and applying gentle pressure.

Leg length

When full knee extension is possible: Position the patient supine on the plinth with the pelvis as straight as possible. The trunk should be straight, head midline, arms by sides. The patient should not lift the head during measuring. Measure from the ASIS (press the end of the tape up against the underside of the ASIS) to the distal end of the medial malleolus.

When significant knee deformity exists: Measure from ASIS to medial malleolus but via the medial condyle. Apply this method to both sides.

Tibial Torsion (transmalleolar axis)

Footprint method:

- Place the foot on a sheet of lined paper, with the knee axis parallel to the lines. The thigh should line up with the long axis of the paper, with the tibial tubercle pointing forward. When the knee is flexed and extended, the foot should stay aligned with the long axis of the paper.
- Draw round the foot
- With a set square, mark a point vertically below the middle of each malleolus.
- Remove the paper and draw a line through the two points marking the malleoli – the ankle axis.
- Measure the angle between the ankle axis and any of the lines on the sheet of paper – this is the angle of the transmalleolar axis.
- External rotation of the foot (most common) is entered as a negative value in Vicon.

Marker Placement

The marker placements are based on the Plug-in-Gait model for Vicon. For details refer to the document ‘Procedure for Marker Placement’ (markerplacement.doc), version 6, 07/03/2011.

Motion Capture

Motion capture is performed using the *plug-in-gait* model in Vicon. A balance test will be performed in addition to level gait. Kinetic data will be recorded using the two AMTI force plates. A minimum of 5 trials for each side of kinematic data should be available for analysis as well as 3 trials of kinetic data.

Static trial

A static trial is recorded for a couple of seconds with the knee alignment devices (KADs) in place. Ensure that all markers are visible in the trial. Remove the KADs and replace with 1 marker on the lateral knee joint centre.

Walking trial

The participants will be asked to look straight ahead and walk at a comfortable pace across the lab. The starting position should be adjusted to allow the force plates to be struck properly. The kinetic data can only be analysed properly if one foot, and only one foot, lands completely on the force plate. The participants should not be made aware of the force plates since this could lead to targeting and altered gait patterns.

Gross Motor Function Measure

The Gross Motor Function Measure (GMFM) will be evaluated for dimensions D and E. The scoring used will be that used for the GMFM-66 (with only 37 of 66 items tested), which involves using the Gross Motor Ability Estimator (GMAE) software.

Scoring of the GMFM is detailed in the user's manual (Russell D et al. 2002). Training on scoring of the measure should be completed using the training CD that accompanies the manual (Gross Motor Function Measure (GMFM) Self-Instructional Training CD-ROM).

Shuttle Run Test

The shuttle run test used will be dependent on the GMFCS of the participant. Those at GMFCS I and II will run between two marks that are 10 m apart; however, there is a different audio CD for the bleeps in each case. Those at GMFCS I begin at 5 km/h with an increase of 0.25 km/h approximately every minute. Those at GMFCS II begin at 2 km/hour with an increase of 0.25 km/h approximately every minute.

The participants with a GMFCS III will use the same audio signals as those at GMFCS II; however, they will run along the sides of a 7.5-metre square (thus requiring only a 90° turn and not 180° turn at each bleep).

During the shuttle run test the participants will walk/jog/run between marks on the floor. They should arrive at the end of a 'shuttle' by the time the audio CD gives a bleep. For each level the time between bleeps is reduced and the participants must increase their speed accordingly. If the participants are having trouble understanding the concept or adjusting their speed, the researchers should run the shuttles with them until they are comfortable with the concept. The shuttle run test should be continued until the participants are too tired to keep up with the pace of the beeps. The participants are given the opportunity to 'catch up' on the next bleep if they fail to reach the end of one shuttle in time for the bleep. However, the test ends when two shuttles are missed in a row.

ActivPAL™

At the end of the assessment the participants should be provided with an ActivPAL™ unit to wear for 7 days. Each participant should be told how to use the ActivPAL™ and given written instructions on its use. They should also be provided with a padded envelope in which to return the unit after the 7 days are complete.

7.4.8 Outcome measures

Physical function was measured with the TUG, 10m SRT and isometric muscle strength (hip extensors, hip abductors and knee extensor), GMFM 66 dimensions D & E, COPM and FAQ. Gait quality was assessed by calculating the gait profile score (GPS) derived from the three-dimensional gait analysis (3DGA) gait kinematics. Seven healthy controls were recruited by CS (previous research assistant responsible for the pilot study of the CP exercise study). The mean age of the healthy controls was 23.6 years and the standard deviation was 0.89. Objective habitual physical activity was measured using the activity monitor (ActivPAL^{TM}). To measure QoL, the Short Form 12 (SF-12) version 2 was used. The RSES was administered to assess the participants' self-esteem. The reader is referred to Chapter 3 (General Methodology) for further details relating to each outcome measure.

In addition, at the end of the 18 weeks, the participants in the exercise group were asked to complete a questionnaire exploring their perception of several characteristics of the exercise programme (e.g. frequency of the exercise sessions, level of difficulty, variation, etc.), as well as information on their current exercise or physical activity routine (Appendix 12).

7.4.9 Statistical analysis

All analyses were performed using SPSS version 21 (SPSS Inc, Chicago, IL, USA). A significance level of $p<0.05$ was used for all tests. The following analyses were performed:

a) to determine any differences between the exercise and control groups at baseline, Mann-Whitney and Fisher exact tests were used as appropriate.
b) a repeated measure of the General Linear Model Analysis of Variance (ANOVA) was performed to detect a possible interaction effect between time and group, i.e. for research questions 1 and 2.

c) a non-parametric Friedman test was performed to investigate, in the intervention group alone, any within-group changes for the outcomes of physical function and gait quality at 6, 12 and 18 weeks, i.e. for research question 3.
d) the Wilcoxon Signed-Rank test was performed to investigate the outcomes of objective habitual physical activity patterns, self-reported function (COPM, FAQ), QoL and self-esteem in the exercise group changes from baseline to 12 weeks (i.e. for research question 4).

For the habitual physical activity data derived from the ActivPAL^{TM}, the participants' data were only included for analysis if at least four full days (24 hours each) of data were available. The data were visually inspected and from this it was subjectively judged whether the ActivPAL^{TM} was worn or not.

When a significant effect over time was identified with analysis c, a post-hoc test (Wilcoxon Signed-Rank) was performed with a Bonferroni adjustment with the level of significance set as $p<0.017$ (comparisons of three situations: baseline–6 weeks, baseline–12 weeks, baseline–18 weeks).

Cohen's *d* values were calculated to obtain the effect sizes of the exercise programme at each time point (6 weeks, 12, weeks, 18 weeks) in the experimental group (Cohen 1988) and interaction groups x time at week 6 for physical function and gait quality and at 12 weeks for self-reported function, habitual physical activity, QoL and self-esteem. Effect size (*d*) was regarded as 'small' if $d = 0.2–0.49$, 'moderate' if $d = 0.5–0.79$, and 'large' if $d > 0.8$ and was calculated using the following formula (Cohen 1988):

$$d = \frac{M_1 - M_2}{SD_{pooled}}$$

Where M_1 and M_2 are the means of intended outcome measure (i.e. TUG) for the experimental and control groups, respectively, and SD_{pooled} is the pooled

standard deviation for the samples (intended outcome measure). SD_{pooled} is calculated using this formula:

$$SD_{pooled} = \sqrt{\frac{SD_1^2 + SD_2^2}{2}}$$

Where SD_1 and SD_2 represent the standard deviations for the experimental and control groups, respectively.

7.5 RESULTS

7.5.1 Participants

Four hundred and thirty young people with CP aged 16 to 25 years old were sent information about the study and were invited to take part, which was carried out in two batches approximately 1.5 years apart (Figure 7.3). The duration of the participants' recruitment for both batches was approximately 1.5 years. In total, 28 individuals returned the form expressing their interest in taking part. Of those 28, one was not suitable for inclusion as he was already following a gym-based exercise programme, and eight changed their minds or did not attend for the initial assessment.

Nineteen participants gave informed consent at their baseline assessment and were randomly allocated to either the exercise (experimental) or control group. At the week 6 assessment, one participant from the control group discontinued participation in the programme. After the 6-week assessment, one participant in the control group wished to join in the exercise programme. After week 12, two participants in the experimental group withdrew from the programme due to unrelated existing health problems (one participant due to high anxiety/depression and another participant due to chronic fatigue syndrome). Three participants in the control group subsequently enrolled in the exercise programme after the 12-week assessment, thus leaving two participants in the

control group for the period of weeks 12–18. At week 18, one participant in the control group did not attend the assessment.

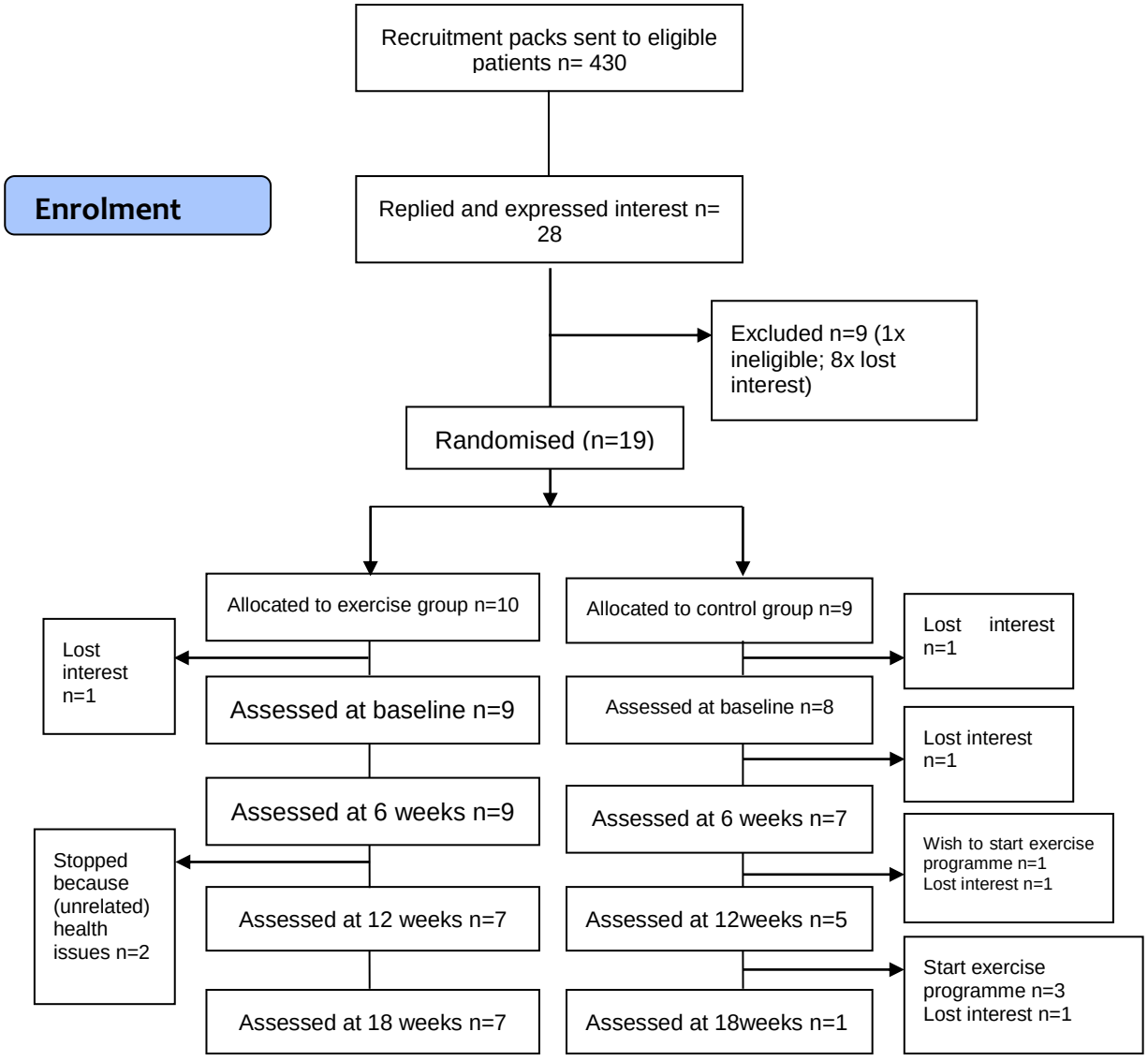


Figure 7.3 Flow chart of participants through the 18-week study period

Over the course of the study there were missing data for some outcome measures. If the assessor (CS or AZ) deemed it clinically unsafe to perform the outcome measure (e.g. hip extensor and abductor strength test aggravating

back pain), then the test was not completed and it was recorded as missing data. This occurred on two occasions with participant (P994) with GMFCS level I. One participant in the exercise group was unable to perform the 10m SRT and data for one participant, 3DGA, could not be recorded as his walking frame obscured and knocked off some of the reflective markers. One participant was not able to perform the 10m SRT.

7.5.3 Descriptive results

Table 7.3 provides the demographic data for each group – experimental and control – for which at least two assessments (i.e. baseline and 6 weeks) were available. The age, height, body mass, gender distribution and means of all outcome measures were similar in the two groups (Table 7.4) at baseline.

Table 7.3 Demographics of the participants at baseline in Study 3

	Experimental group	**Control group**
Number of participants	9	7
Gender M:F	3:6	3:4
Age (years)	20.0(3.0)	20.0(3.2)
Body mass (kg)	61.3(22.8)	62.8(18.3)
Height (m)	1.62(9.3)	1.61(13.3)
GMFCS I:II:III	4:2:3	2:3:2
Diagnosis	Hemiplegia: 2 Diplegia: 6 Ataxia: 1	Hemiplegia: 3 Diplegia:3 Tetraplegia:1
Walking aids		
No walking aid	6	6
Canes	1	-
Walker	2	1

M:males; F:females; kg: kilogrammes; cm:centimetres; GMFCS: Gross Motor Function Classification Score

Table 7.4 Comparison of baseline measures between groups

	Experimental n=9	Control n=7	p-value*
Gender (M:F)	3:6	3:4	0.55
Age (years)	20.04(3.0)	19.97(3.2)	1.00
TUG(s)	19.0(12.6)	18.5(12.7)	0.87
10m SRT	8.7(6)	8.7(4)	0.86
Hip extension (Nm/kg)	1.04(0.5)	1.32(0.6)	0.25
Hip abduction (Nm/kg)	0.49(0.2)	0.57(0.3)	0.63
Knee extension (Nm/kg)	0.85(0.3)	0.85(0.4)	0.87
GMFM D & E	69.5(15.9)	70.0(13.4)	0.56
GPS	15.66(5.7)	13.7(4.9)	0.56
Sitting/lying (hour/day)	20.90(1.2)	21.49(1.4)	0.49
Standing (hour/day)	2.09(0.7)	1.78(1.1)	0.49
Steps/day	3587(3532)	3300(2038)	1.0
Sit to stand (no/day)	39.75(17.9)	35.29(11.8)	0.91
COPM (performance 0-10)	3.6(2.1)	3.9(1.2)	0.57
FAQ (0-25)	15(7)	16(9)	0.67
PCS (34.75-56.71)	42.72(10.1)	42.40(8.0)	0.87
MCS (15.37-62.39)	41.76(15.6)	49.71(10.7)	0.22
RSES	16.56(7.3)	23.0(3.7)	0.06

**All tested with Mann-Whitney test except for gender for which the Fisher Exact was used TUG: Timed Up and Go test; s:seconds; 10m SRT:10-metre shuttle run test; Nm/kg: NewtonMetre/kilogramme; GMFM D & E: Gross Motor Function Measure Dimensions D &E; GPS: Gait Profile Score; COPM: Canadian Occupational Performance Measure; FAQ: Gillette Functional Assessment Questionnaire; PCS:Physical Component Score; MCS: Mental Component Score; RSES: Rosenberg Self-Esteem Scale*

7.5.2 Feasibility aspects of the study: adherence, adverse events, missing data and feedback regarding the exercise programme

7.5.2.1 Adherence

Exercise session attendance was recorded for the experimental group at week 6 (Block 1), week 12 (Block 2) and week 18 (Block 3). Overall adherence in Block

1 was 86% (124 out of a possible 144 sessions). For Block 2, the adherence was 90.6% (87 out of the minimally prescribed number of 96 sessions) (Table 7.5). Interestingly, only two participants (P15, P17) exercised thrice weekly in Block 2, and this dropped to once or twice a week in Block 3. There was no prescribed minimal exercise frequency for Block 3, but the participants exercised on average once per week (ranging from less than once a week to twice a week).

Table 7.5 Number of exercise sessions as a proportion of the prescribed number of sessions for each block of six weeks

	Block 1	Block 2	Block 3	Total
	Week 1–6[a]	Week 7–12[b]	Week 13–18[c]	
P449	16	13	8	37
P848	18	5	5	28
P318	18	12	12	42
P994	n/a	n/a	n/a	n/a
P236	18	6	2	26
P11	10	0	0	10
P12	11	15	12	38
P15	18	18	8	44
P17	15	18	11	44
Total	124/144 (86%)	87/96 (90.6%)	58	

n/a – not available (participant did not return their exercise logbook)
[a] *three prescribed sessions per week with 18 possible sessions for the whole of Block 1*
[b] *two prescribed sessions per week with 12 possible sessions for the whole of Block 2*
[c] *self-directed/unsupervised exercise sessions*
144 sessions= 8 (participants) x 6 (weeks) x 3 (sessions/week)

7.5.2.2 Adverse effects

There were no serious adverse effects during the exercise programme apart from minor musculoskeletal aches and general fatigue claimed by one participant (P15 in one session). P236 commented in their logbook regarding

tiredness during nearly all of the sessions from week two onwards; however, P236 attended 100% of the sessions for the first block and for two weeks of the thrice-weekly sessions in the second block prior to withdrawing from the programme due to chronic fatigue syndrome.

7.5.2.3 Feedback on the exercise programme

All nine of the participants in the exercise group completed the feedback questionnaire (Table 7.6). All of the participants considered the length of the session (one hour) to be just right and would have liked to continue the programme. However, opinions on the content of the programme varied. Five of the participants found the programme to at times be too easy, while three found it too hard at times. Two did not like the fact there were no changes in the content of the programme and would have liked more variety to be introduced, but the others (n=7) liked the fact there were no substantial changes.

Table 7.6 Feedback on exercise programme questionnaire results

	Level of exercises	Group or individual	Length of session	Variety of exercises	Alterations to the programme	Adverse effects	Amount of supervision/ instruction	Location of exercise	Benefits noted	Would you continue with the exercise programme?	If, so which components?	And where?	Other physical activity?	Start any new activities?	Other comments?
P339	Usually but sometimes too easy	Makes no difference	Just right	Good variety of exercises	There were no alterations and I liked that	At the start I had sore legs	Just right, the instructors were very good	Local leisure centre	Transferring was easier	Yes, but only for 1-2 sessions a week	Strength, aerobic	I'd like to continue going to my local leisure centre	Boccia	No	N/A
P848	Usually but sometimes too easy	Alone	Just right	Not enough variety	There were no alterations and I liked that	No	Not enough, instructors hardly in the gym	Local leisure centre	Results were positive as muscles become looser and range of motion better, also lost some weight	Yes	Strength, stretching	I'd like to continue going to my local leisure centre	Baseball, swimming	no	Exercises too repetitive, not enough variety
P449	Usually but sometimes too easy	Group	Just right	Not enough variety	There were no alterations and I liked that	No	Just right	Local leisure centre	Feel stronger and fitter	Will change to a different programme	Strength, aerobic	I'd like to continue going to my local leisure centre	Horse riding, swimming	No	I did not find it varied enough and I would like the progamme to change every 4 weeks
P236	Usually but sometimes too difficult	Group	Just right	Good variety of exercises	I liked how the sessions were altered as I got more used to the exercises	Felt dizzy and tired after gym, it was bearable but as we went on it got worse	Just right, Gill was a great support	Local leisure centre, Meadowbank not a good venue	Feel weaker but felt that is Post Viral Fatigue Syndrome	Feel weaker but felt that is PVFS	stretching	I will modify the programme to do as much as possible at home	Swimming, fencing (only 1/week as very tired)	No	I found it difficult to take part in this, wanted to continue with it but was having severe pain, tiredness and depression

	Level of exercises	Group or individual	Length of session	Variety of exercises	Alterations to the programme	Adverse effects	Amount of supervision/ instruction	Location of exercise	Benefits noted	Would you continue with the exercise programme?	If, so which components?	And where?	Other physical activity?	Start any new activities?	Other comments?
P318	Usually but sometimes too easy	Makes no difference	Just right	Good variety of exercises	There were no alterations and I liked that	Yes, on the double leg push. My hamstrings became much tighter than usual	Just right	Own health club, staff knew me and understood my needs	Not really, core stability and balance maybe slightly	Yes but only 1-2 a week	Strength, aerobic	Continue at health club	Swimming, static bike	No	N/A
P11	Usually but sometimes too difficult	Alone	Just right	Good variety of exercises	N/A	Muscle pains	Just right	Local leisure centre	Easier to move, felt less breathless	N/A	All components	I'd like to continue going to my local leisure centre	N/A	No	N/A
P12	Usually but sometimes too easy	Group	Just right	Good variety of exercises	I liked how the sessions were altered as I got more used to the exercises	I had minor knee pain in my first few sessions with the program	Just right	Local leisure centre	Increase in stamina and found it easier cycling	Yes	All components	Other: I got a free gym membership from college	Cycling, squatting	Kung fu, swimming	Had a great time participating in this program. Thanks ☺
P15	Usually but sometimes too difficult	Group	Just right	Good variety of exercises	I liked how the sessions were altered as I got more used to the exercises	Muscle soreness on beginning	Just right	Local leisure centre	N/A	Yes but only 1-2 a week	All components	I'd like to continue going to my local leisure centre	Stretching	No	This program was a good idea, was feeling motivated, to keep fit , more happier
P17	Yes	Group	Just right	Good variety of exercises	I liked how the sessions were altered as I got more used to the exercises	Not applicable	Just right	I'm a member of Gullane Gym and like going there	Yes. Lost some weight. More mobile. A bit steadier in exercises	Yes but only 1-2 a week	Strength, stretching	Other: Gullane gym	Swimming	No	The program lasted longer than I expected, and became a little difficult to fit in to my routine

7.5.4 The outcomes of physical function in the intervention group after 6 weeks compared to those in a control group

The repeated-measures ANOVA test showed no statistically significant interaction group x time effects at 6 weeks in any of the physical function measures. The TUG showed a statistically significant time effect, which indicated that at 6 weeks both groups took less time to complete the TUG. The GMFM scores also show a statistically significant increase over time (6 weeks). The p-values presented in Table 7.8 are based on the participants who had data for both sessions, as presented in Table 7.7.

Table 7.7 Results of the physical function and gait quality measures after the exercise programme (6 weeks)

Outcome measures		Baseline (mean)		6 weeks (mean)	
		Exercise n=9	Control n=7	Exercise n=9	Control n=7
Physical function	TUG(s)	19.0(12.1)	18.5(12.7)	16.7(12.2)	15.8(9.9)
	10m SRT	8.7(6)	8.7(4)	9.0(6)	9.0(3)
	Hip extension (Nm/kg)	1.04(0.46)	1.32(0.55)	1.09(0.55)	1.52(0.7)
	Hip abduction (Nm/kg)	0.49(0.22)	0.57(0.31)	0.58(0.24)	0.58(0.28)
	Knee extension (Nm/kg)	0.85(0.29)	0.85(0.29)	1.12(0.42)	1.02(0.51)
	GMFM D & E	69.5(15.9)	70.0(13.4)	71.5(16.4)	70.8(13.8)
Gait quality	GPS (º)	15.66(5.7)	13.7(4.9)	15.79(5.6)	15.06(5.5)

TUG: Timed Up and Go test; s:seconds; 10m SRT: 10-metre shuttle run test; Nm/kg: NewtonMetre/kilogramme; GMFM D & E: Gross Motor Function Measure Dimensions D & E; GPS: Gait Profile Score

Table 7.8 Differences between groups and over time (6 weeks)

Outcome measure		Group effect p-value	Time effect p-value	Group/time interaction p-value	Partial Eta Squared[a]	Cohen's *d*[a]
Physical function	TUG	0.86	0.03*	0.57	0.02	0.03
	10m SRT	0.99	0.49	0.93	0.01	0.00
	Hip extension	0.22	0.21	0.44	0.16	0.14
	Hip abduction	0.77	0.37	0.43	0.05	0.18
	Knee extension	0.77	0.06	0.64	0.02	0.14
	GMFM D & E	0.99	0.05	0.33	0.07	0.02
Gait quality	GPS	0.64	0.39	0.48	0.00	0.11

**significant difference over time p<0.03*
[a] Group/time interaction
TUG: Timed Up and Go test; 10m SRT: 10-meter shuttle run test; GMFM D & E: Gross Motor Function Measure Dimensions D & E; GPS: Gait Profile Score

7.5.5 The outcomes of objective habitual physical activity, self-reported function (COPM, FAQ), QoL and self-esteem in the exercise group after 12 weeks compared to those in a control group

There was no statistically significant group x time interaction effect at 12 weeks in the habitual physical activity, QoL and self-esteem measures. At week 12, the self-reported function measured by the COPM increased significantly in both groups with p=0.02. The statistical results shown in Table 7.10 are based on those participants who had data for both sessions, as presented in Table 7.9. The list of activities selected and scored by the participants as those they would

like to improve through following the exercise programme (using the COPM) is presented in Table 7.11.

Table 7.9 Results of physical activity, self-reported function, QoL and self-esteem after the exercise programme (12 weeks)

Outcome measure		Baseline (mean)		12 weeks (mean)	
		Exercise	Control	Exercise	Control
HPA	Sitting/lying* (hour/day)	20.8(1.4), n=5	21.2(1.6), n=5	20.2(2.2), n=5	20.5(1.7), n=5
	Standing* (hour/day	2.2(0.9), n=5	1.8(1.0), n=5	2.5(1.3), n=5	2.5(1.2), n=5
	Steps/day*	4573(4270), n=5	3883(2138), n=5	5357(4625), n=5	4998(2628), n=5
	Sit to stand*	43 (22), n=5	35(12), n=5	43(14), n=5	37(11), n=5
Self-reported function	COPM (Performance 0-10)	3.6(2.1), n=7	3.9(1.2), n=5	5.1(2.0), n=7	4.5(1.1), n=5
	FAQ (0-25)	15(7), n=7	16(9), n=3	16(5), n=7	16(9), n=3
QoL	PCS (34.75-56.71)	46.2(9.3), n=6	44.8(7.9), n=5	44.9(8.2), n=6	43.4(11.1), n=5
	MCS (15.37-62.39)	47.5(12.7), n=6	52.2(10.9), n=5	53.0(14.1), n=6	53.7(11.7), n=5
Self-esteem	RSES (0-30)	18.4(8), n=5	23.6(4), n=5	19.2(9.7), n=5	24.6(2.6),n=5

HPA: habitual physical activity; COPM: Canadian Occupational Performance Measure; FAQ: Gillette Functional Assessment Questionnaire; QoL: Quality of life; PCS:Physical Component Score; MCS: Mental Component Score; RSES: Rosenberg Self-Esteem Scale
**derived from four days' data*

Table 7.10 Differences between groups and over time (week 12) for physical activity, QoL and self-esteem

Outcome measure		Group effect p-value	Time effect p-value	Group/time interaction p-value	Partial Eta Squared[a]	Cohen's *d*[a]
HPA	Sitting/lying	0.77	0.09	0.86	0.00	0.06
	Standing	0.94	0.13	0.77	0.01	0.36
	Steps/day	0.82	0.12	0.77	0.01	0.09
	Sit to stand	0.43	0.90	0.82	0.00	0.13
Self-reported function	COPM	0.91	0.02*	0.33	0.10	0.54
	FAQ		0.94	0.33	0.12	0.13
QoL	PCS	0.26	0.26	0.22	0.00	0.01
	MCS	0.94	0.17	0.58	0.11	0.32
Self-esteem	RSES	0.37	0.62	0.47	0.00	0.03

**significant difference over time p<0.05*
[a] group/time interaction
HPA: habitual physical activity; COPM: Canadian Occupational Performance Measure; FAQ: Gillette Functional Assessment Questionnaire; QoL: Quality of life; PCS:Physical Component Score; MCS: Mental Component Score; RSES: Rosenberg Self-Esteem Scale

Table 7.11 List of activities derived from the Canadian Occupational Performance Measure from baseline to week 12

		Activity (ies)	Performance score (0-10)	
			Baseline	12 weeks
Exercise group	P848	Balance (drying off from shower)	5	5
		Long distance walking	4	5
		Going up stairs	5	5
		Up/down curbs on own	1	1
		Run	3	3
	P449	Carrying drinks	5	5
		Stairs without a railing	6	7
		Escalators	6	6
		Uneven ground/balance	5	6
		Stamina over long distances	7	8
	P318	Carrying liquids	1	1
		Standing without hands	3	4
		Escalators	1	1
		Down spiral staircase	2	2
		Dressing in kilt	2	2
	P11	Swimming	2	2
		Walk less bouncy	1	3
		Walk more than 5 minutes	1	5
		Right hand function	4	4
	P12	Climbing (indoor)	5	8
		Cycling	7	7
		Sailing	8	8
	P15	More walking without walker	1	6
	P17	Running (quicker)	5	7
		Swimming (kicking)	0	2
		Climb down from stairs (safer)	7	9
Control group	P339	Dressing	8	8
		Transferring chair to walker	7	8
		Holding objects when standing	2	2
		Posture when walking	2	2
		Confidence to take steps without aids	3	2
	P831	Carrying heavy objects	4	6
		Carrying unbalanced objects	3	7
		In/out of tub	8	8
		Carrying liquids	6	5
		Muscle endurance/stamina	6	4
	P659	Bath	1	1
		Escalators	1	2
		Long walks	4	5
		Dancing	3	4
	P16	Cycling (starting)	2	2
		Kneeling	3	3
		Fitness level (more energy)	4	5
		Upper limb strength	3	3
	P210	Two-wheel bicycle	1	1
		Balance	5	6
		Carrying objects	6	7
		Swimming	3	3
		Walking long distance	6	7
	P14	Walking (distance)	2	2
		Horse riding	5	7
		Ride bike	1	1

7.5.6 The outcomes of physical function in the exercise group over time (6, 12, 18 weeks)

As only a very small number of the participants in the control group attended for assessment at weeks 12 and 18, analysis on the effects over time (6, 12, 18 weeks) was performed for the exercise group only. The physical function and gait quality data for the exercise group over time are presented in Table 7.12. There was a significant difference in hip extension strength over time in the exercise group. However, post-hoc tests with Bonferroni adjustment ($p>0.017$) revealed that no significant change occurred between baseline and 6 weeks ($p=0.89$, $d=0.14$), baseline and 12 weeks ($p=0.09$, $d=0.76$) or baseline and 18 weeks ($p=0.043$, $d=0.41$). There were no statistically significant differences over time in other measures in the experimental group (Table 7.13). Hip abductor strength showed a continual increase from baseline to week 12, with a moderate effect size ($d=0.54$). Moderate effect sizes were also found for knee extensor strength at week 6 and week 18, with $d=0.67$ and $d=0.79$, respectively.

Table 7.12 The outcome (mean) of physical function and gait quality over time in the experimental group

Outcome measure	Baseline n=7	Week 6 n=7	Week 12 n=7	Week 18 n=7
TUG(s)	17.1(11.4)	15.8(11.1)	15.0(9.8)	16.8(13.3)
10m SRT	9.1(6.4)	9.4(6.4)	8.3(5.3)	8.8(6.7)
Hip extension (Nm/kg)	1.04(0.49)	0.97(0.48)	1.46(0.74)	1.28(0.67)
Hip abduction (Nm/kg)	0.59(0.16)	0.67(0.20)	0.69(0.21)	0.68(0.27)
Knee extension (Nm/kg)	0.86(0.26)	1.1(0.45)	1.0(0.4)	1.1(0.34)
GMFM D & E	70.5(16.4)	72.7(17.0)	72.2(17.9)	72.4(17.6)
GPS (^{o})	15.66(5.7)	15.79(5.6)	15.78(4.4)	14.33(3.1)

TUG: Timed Up and Go test; s:seconds; 10m SRT: 10-metre shuttle run test; Nm/kg:NewtonMetre/kilogramme; GMFM D & E: Gross Motor Function Measure Dimensions D &E; GPS: Gait Profile Score

Table 7.13 Differences over time (baseline, week 6, week 12, week 18) for physical function in the experimental group (Friedman test)

Outcome measure		p-value	Post hoc*	Effect size		
				Week 6	Week 12	Week 18
Physical function	TUG	0.18	n/a	0.12	0.2	0.02
	10m SRT	0.41	n/a	0.05	0.14	0.05
	Hip extension	0.03	p=0.89[a],0.09[b],0.043[c]	0.14	0.67	0.41
	Hip abduction	0.49	n/a	0.44	0.54	0.41
	Knee extension	0.62	n/a	0.65	0.42	0.79
	GMFM D & E	0.50	n/a	0.13	0.10	0.11
Gait quality	GPS	0.62	n/a	0.02	0.02	0.29

**Bonferroni adjustment significant value =p<0.017*; *TUG: Timed Up and Go test;10m SRT: 10-metre shuttle run test; GMFM D & E: Gross Motor Function Measure Dimensions D &E; GPS: Gait Profile Score*
[a] p-value between baseline and 6 weeks with Bonferroni adjustment (Wilcoxon Ranked Test)
[b] p-value between baseline and 12 weeks with Bonferroni adjustment (Wilcoxon Ranked Test)
[c] p-value between baseline and 18 weeks with Bonferroni adjustment (Wilcoxon Ranked Test)

7.5.7 The outcomes of objective habitual physical activity patterns, self-reported function, QoL and self-esteem in the exercise group: changes over time (12 weeks)

The outcomes for objective habitual physical activity, self-reported function, QoL and self-esteem were explored over time from baseline to week 12 in the exercise group and are shown in Table 7.14. There was a statistically significant effect of time for self-reported function, as measured by COPM (Table 7.15). The current study also found a trend (*d*=0.36) of improvement in the exercise group for time spent sitting/lying and standing using data derived from at least 4 days' use of the ActivPAL^{TM} from baseline to week 12, but this did not reach statistical significance (p=0.3 & 0.5). At week 12, the mean time for sitting/lying (sedentary behaviour) was reduced by 0.6 hours and the number of steps per day had increased by 949 steps; however, these changes were less than the MDC values found in Study 2, Chapter 5, thereby indicating that the changes could be due to measurement error. Although no statistical effects of time were found in the mental component score (MCS), a trend of improvement was observed with a small effect size (*d*=0.34).

Table 7.14 The outcome (mean) of the objectives habitual physical activity, QoL and self-esteem over time in the experimental group

Outcome measure		Baseline	Week 12
HPA	Sitting/lying (hour/day)	20.9(1.4), n=5	20.3(1.9), n=5
	Standing (hour/day)	2.1(1.0), n=5	2.5(1.2), n=5
	Steps/day	4228(3204), n=5	5177(3552), n=5
	Sit-to-stand transition	39(17), n=5	40(12), n=5
Self-reported function	COPM (Performance 0-10)	2.8(1.9), n=7	4.3(1.8), n=7
	FAQ (0-25)	14.6(5.7), n=7	15.6(3.9), n=7
QoL	PCS (34.75-56.71)	44.1(8.7), n=7	42.1(5.4), n=7
	MCS (15.37-62.39)	48.1(14.0), n=7	53.2(15.8), n=7
Self-esteem	RSES	18.4(8.4), n=7	19.2(9.7), n=7

HPA: habitual physical activity; COPM: Canadian Occupational Performance Measure; FAQ: Gillette Functional Assessment Questionnaire; QoL: Quality of life; PCS:Physical Component Score; MCS: Mental Component Score; RSES: Rosenberg Self-Esteem Scale

Table 7.15 Differences over time (baseline–week 12) for physical activity, QoL and self-esteem in the experimental group (Wilcoxon Rank test)

Outcome measure		p-value	Effect size
HPA	Sitting/lying	0.35	0.36
	Standing	0.50	0.36
	Steps/day	0.50	0.28
	Sit to stand	0.89	0.07
Self-reported function	COPM	0.02*	0.81
	FAQ	0.60	0.20
QoL	PCS	0.75	0.28
	MCS	0.08	0.34
Self-esteem	RSES	0.21	0.09

**significant difference over time p<0.05; HPA: habitual physical activity; COPM: Canadian Occupational Performance Measure; FAQ: Gillette Functional Assessment Questionnaire; QoL: Quality of life; PCS: Physical Component Score; MCS: Mental Component Score; RSES: Rosenberg Self-Esteem Scale*

7.6 Discussion

7.6.1 Analysis of group x time interaction at 6 weeks (RCT)

This analysis aimed to compare the results for physical function (TUG, 10m SRT, isometric muscle strength, GMFM, COPM, FAQ) and gait quality between the intervention (exercise) group following 6 weeks of a pragmatic community exercise programme and a control (usual care) group. The results showed no significant time x group interaction at 6 weeks for any of the outcome measures. Other RCTs that examined the effects of exercise interventions for people with CP also failed to reveal between-group differences for some or all of the outcome measures (Dodd et al. 2003, Taylor et al. 2013). Dodd et al. (2003) reported on the results of an RCT into the effects of a six-week home-based strength training programme involving 21 participants with CP aged 8 to 18 years classified as GMFCS levels I–III, and they also did not find a significant group x time interaction for either isometric muscle strength of the hip extensors and abductors or for the GMFM dimensions D&E. However, Dodd et al. (2003) did report that the exercise group showed a statistically significant increase in

combined ankle plantarflexor and knee extensor strength at 6 and 12 weeks compared to the control group. Another RCT on 12-week strength training also failed to find a difference (i.e. improvement) on GPS and GMFM dimensions D & E between the exercise and control groups at the 12-week assessment on participants with CP and a mean age of 18.1 years with GMFCS levels II & III (Taylor et al. 2013). However, Taylor et al. (2013) did report that the exercise group significantly improved on their FAQ, Functional Mobility Scale (at 5m) and the strength of their targeted muscle group compared to the control group.

The highest effect size found in the RCT analysis in this study was for the COPM, which indicated a moderate effect size of *d*=0.54 at 12 weeks. Post-hoc power analysis revealed that if this effect size of 0.54 were to be maintained, a sample size of n=51 in each group would be required for this comparison to reach statistical significance (at 80% probability).

7.6.2 Analysis of the effect of time at 6 weeks (both groups)

This analysis revealed a statistically significant improvement over time in the physical function of both groups, as measured by the TUG, and gross motor function, as recorded by GMFM 66 dimensions D & E. These results suggest that the participants in both groups may have experienced a learning effect in these outcome measures.

7.6.3 Analysis of the exercise group (Within group) at 6, 12 and 18 weeks

The current study found no significant improvement over time (6, 12, 18 weeks) in any of the outcome measures within the exercise group. Some of the OMs – hip extension strength (*d*=0.67) and hip abductor strength (*d*=0.54) – showed moderate effect sizes for improvement from baseline to week 12. However, hip extension strength decreased from weeks 12 to 18 (*d*=0.41). The changes in the scores appear to be different from the results of the significance testing due to

the small sample size. The change in hip extensor strength showed an increase from baseline to week 12; however, this change was not statistically significant, thus indicating a type I error.

The lack of any statistically significant improvement in isometric muscle strength within the experimental group found in the current study is in contrast to the findings reported by Eek et al. (2008), who found a significant improvement compared to baseline for hip extension and hip abductor strength in participants with CP aged 9 to 15 years with GMFCS levels I & II following an 8-week strength-training programme carried out three times per week. This discrepancy in the results may be due to the fact that Eek et al. (2008) focused on strength training only, while the current study combined both aerobic and strength training.

There was a lack of effect of the exercise programme on the outcomes of walking performance and gait quality. The TUG test and gait quality measured by the GPS did not show improvement in any of the assessments (6, 12 and 18 weeks), as evidenced by the lack of statistical significance and low effect sizes ($d<0.3$). Other CP studies with adolescents reported mixed effects of exercise programmes on walking speed and gait kinematics. Unger et al. (2008), in an evaluation of an individualised strength programme for 13–18-year-olds, reported a decrease in crouch gait but no effect on walking speed and stride length. In a different study by Darrah et al. (1995) with participants aged 11–20 years, significant improvements in strength were reported but no significant change in walking speed.

Although there was a clear lack of change in objective physical function outcomes over time, there were statistically significant improvements in self-reported physical function, as measured by the COPM, from baseline to week 12 in the experimental group, with large effect size ($p=0.02$, $d=0.81$). The COPM

is a self-reported questionnaire in which participants are asked to list those activities (a maximum of five) that they would like to improve by following the exercise programme. The participants in the exercise group rated improvement in the performance of their selected daily activities, such as distance walked, climbing up and down the stairs and swimming, from baseline to week 12 by nearly one point more (from 3.6 to 5.1) compared to the control group (3.9 to 4.5). This result indicates that the exercise programme influenced the individuals' rated performance regarding their performance in their daily activities, which perhaps allowed them to participate more in society (McBurney et al. 2003). However, the COPM is a very subjective outcome measure and thus the positive result could be (partly) due to the fact that the participants anticipated an improvement through their following of the exercise programme.

There was no significant difference observed either within group (experimental) or between groups with regards to the FAQ scores from baseline to 12 weeks in this study. The current finding is consistent with the lack of change reported by McNee et al. (2009) in a study of 13 participants with CP aged 6–16 years who followed a 10-week plantarflexor strengthening programme. It is possible that the FAQ is not sufficiently sensitive to capture relatively small improvements in walking function.

There was a trend for improvement in the exercise group in comparison with the control on MCS scores over the 12 weeks, with a small effect size (d=0.32). Slaman et al. (2014b) found a significant improvement in the MCS (SF36) at the week 6 and week 12 assessment following a combination of physical fitness and counselling intervention in 57 young people with CP. Previous CP exercise studies have measured QoL using population-specific QoL scales such as TACQOL (Verschuren et al. 2007) and PedsQoL (Engsberg et al. 2006) and generic scales such as SF 36 (a longer version of SF-12) (Slaman et al. 2014b).

The RSES was used to evaluate changes in self-esteem in the current study, with the current study finding no statistically significant difference between the exercise and control groups at week 12. Dorval et al. (1996), in a study on the impact of a 10-week aquatic programme on adolescents with CP, also revealed no significant difference in self-esteem measured by the RSES in the experimental group (aquatic programme) compared with the control group following the training and nine-month follow-up. There are other examples of exercise programmes having a positive effect on self-perception in CP (Dodd et al. 2004, Unger et al. 2006). High self-esteem can lead to positive qualities such as life satisfaction, positive social adjustment, independence, adaptability and resilience to stress (Biddle et al. 2003) and has therefore emerged as a strong predictor of mental well-being and quality of life (Diener 1994).

7.6.4 Recruitment, adherence, attrition

While RCTs provide the most reliable evidence for evaluating health care interventions (Barton 2000, Sackett et al. 1996), they are often hindered by recruitment difficulties (Lovato et al. 1997). The recruitment rate in the current study was low (6%) (Figure 7.3) and, as a result, did not achieve the sample size required for an appropriately powered trial. The effects of insufficient recruitment include 1) reduced power to detect significant intervention effects (Swanson and Ward 1995), and 2) generalisability of the results (Easterbrook and Matthews 1992). A systematic review suggested two strategies for increasing recruitment in RCTs. Firstly, engaging participants in learning about the health problem being studied and its impact on their health, or else inform participants of the intervention they have been randomised to receive (non-blind trial design) (Caldwell et al. 2010). This strategy has been applied in our recruitment as the information on the exercise programme was embedded in the invitation package. In addition, the invitation package was revised prior to the current study taking place. In particular, the patient information sheet (PIS)

(Appendix 13) was reviewed (informally, unstructured) by several people with CP aged 16–25 who were attending the Craigalbert Centre (Appendix 14), and this informal discussion does not form part of the thesis. These young people commented on several issues, such as the mentioning of pain in the PIS, which may put some potential participants off. Consequently, the PIS was amended after taking into account their written and verbal comments to the Craigalbert Centre's physiotherapist. Secondly, Caldwell et al. (2010) also suggested the use of monetary incentives. Due to financial restrictions, the current study did not offer these incentives to its participants, although travel expenses from the participants' homes to the assessment venue, and vice versa, were covered. In addition, the participants in the exercise group (as well as the participants in the control group who wished to take part in the exercise programme) received five months' membership of the leisure centre with unlimited access. Future multi-centre trials may improve on the number of participants recruited in comparison to single-centre studies such as the current study.

Adherence was monitored using the exercise logbook. It should be noted that adherence to the programme increased from 86% to 90.6% between Block 1 (0–6 weeks) and Block 2 (7–12 weeks), thereby suggesting that the majority of the participants preferred to exercise twice weekly rather than thrice weekly. This was also supported by the feedback questionnaire, which was returned by nine of the participants upon completion of the exercise programme. Four of the participants stated that they would like to continue to exercise on their own after the programme, but only for 1 to 2 sessions per week. On the other hand, three of the participants answered 'yes' but did not specify a frequency. One participant stated they would like to continue exercising with a different programme, and one participant did not answer. Exit/follow-up questionnaires in future studies should obtain more specific feedback regarding the participants' preferred exercise frequency.

As described above, the majority of the participants exercised at the prescribed frequencies in Block 1 (3x/week) and Block 2 (2x/week), indicating good adherence. However, during Block 3, when physiotherapist input was mostly absent, the average frequency of gym attendance decreased to once per week, thus demonstrating that long-term adherence may not have been achieved for five of the participants; one of the participants did not attend the gym at all and four of the participants attended less than 2/3x per week. Adherence to an exercise programme is multifactorial and there are many age-specific (perceived) barriers to attending the gym (Verschuren et al. 2012, Heller et al. 2002, Shields et al. 2012) (refer to Chapter 8 for further discussion on this). For example, in Block 3, one of the participants was on work placement for three months and therefore could not attend gym sessions at all during these weeks.

Apart from adherence, study attrition was another problem within this study. Five of the participants withdrew over the course of the 18 weeks (at different time points, Figure 7.3). Of those five, two participants were in the exercise group and three participants were in the control group. The attrition rate (29%) in this study is comparable with that seen in previous CP exercise studies (McNee et al. 2009). McNee et al. (2009), in a study of 10-week strength training with frequencies of exercise 4 x/week, had 13 participants complete the programme with an attrition rate of 35%. Since this study has an attrition greater than 20%, this indicates that the results have a high risk of bias; as such, the results should be treated with caution (Dumville et al. 2006).

7.6.5 Fidelity

While the frequency, repetitions and progressive nature of the exercises in the training programme could be quantified through the exercise logbook, the participants' fidelity was not formally measured. The frequency and quality (whether the participants did the exercise as outlined) of training should be

captured and linked to the programme outcomes, to provide stronger evidence on the exercise programme (Fortington et al. 2015). Ideally, to maximise the intervention fidelity, it is recommended that an independent assessor observe the exercise session at random intervals and monitor the consistency of the exercise content using a checklist based on the explicit components of the exercise intervention protocol. However, in practice this might not be practical; hence, the current study used the exercise logbook to monitor the number of repetitions and the progressiveness of the exercise.

7.6.6 Exercise progression and preferences and exercise characteristics

The current study aimed to progress the strength training every 2 weeks when the participants were able to comfortably achieve 12 repetitions with a particular resistance. For aerobic exercise, the aim was to progress the intensity and/or duration whilst keeping the Borg Rating of Perceived Exertion (RPE) scale at '13' using a static bicycle, cross trainer, treadmill or rowing machine as appropriate, preferably by the participants themselves in line with advice from an experienced physiotherapist. The entries in the exercise logbooks showed that all of the participants followed the progression as mentioned above. The feedback questionnaire did not record any complaints that either the dosage or intensity of the exercise programme were too high, thus suggesting progression was well tolerated. The results of the feedback questionnaire also showed that a majority (7 of 9) of the participants would like to continue the exercise programme with strengthening, aerobic and stretching components, with another two participants stating they would prefer to continue with the stretching component only. During the programme the exercise bikes were utilised most. One participant utilised the cross trainer and one utilised the Motomed.

7.6.7 Outcome measures

Despite the large number of outcome measures (13) repeated at four time points with between 2 and 3 hours for each visit, the assessment was shown to be

feasible and did not discourage participation for those who volunteered to participate in the study. No complaints were received regarding the duration of the assessment, but this may be due to the fact that the participants wanted to please the researcher and were eager to find out their performance and any improvement they had made. However, the current study did not ask for feedback regarding the length of the assessment in the exit questionnaire (feedback questionnaire), which was focused on the characteristics of the exercise programme and not the study itself.

It is possible, however, that the potential participants who were invited to participate in the study decided not to participate due to the requirement to commit to relatively frequent and long assessments, thus partly resulting in the low recruitment rate. The duration of the assessment in relation to the recruitment rates should therefore also be examined as this study had a very low recruitment rate (6%), and this is something that could be explored in future studies through a survey or follow-up call. The duration of assessment could potentially be reduced in future studies by performing fewer outcome measures during each visit. But, as discussed in Chapters 2 and 5, the psychometric properties of the outcome measures should be important criteria to consider when choosing a tool to measure outcomes following exercise intervention.

7.6.8 Limitations

In the current study, the target sample size was not achieved due to low recruitment rates and attrition of 29%; hence, the study was underpowered and therefore at risk of type-2 error. As discussed above, the low number of participants may have been due to the requirement to commit to an 18-week, thrice-/twice-weekly exercise programme and the high number of outcome measures assessed (2–3 hour assessments, 4 times). Interestingly, the number of participants in the current study is comparable to those in many similar

studies (Damiano et al. 1995a, McPhail and Kramer 1995, Dodd et al. 2004, Eagleton et al. 2004, Engsberg et al. 2006, Unnithan et al. 2007, McBurney et al. 2003, McNee et al. 2009, Eek et al. 2008, Auld et al. 2014), thus indicating that small sample size is a common problem in exercise studies in the CP population, which is something that needs to be addressed.

The fidelity of the exercise programme was not formally assessed over the duration of the programme. As discussed earlier, having an independent assessor to assess whether the programme was conducted as planned (i.e. progression of the exercise, RPE) would have been preferable. However, the aim of this study was to assess the effects of a pragmatic community exercise programme and not those of a highly structured and supervised programme in a university or hospital setting.

7.7 Conclusion

The RCT did not result in any statistically significant findings; however, moderate effect was found on the COPM, thereby indicating that there was a trend of improvement from baseline to 12 weeks in self-reported physical function. Analysis within the experimental group detected a statistically significant improvement on the COPM at 12 weeks. Moderate effect sizes indicating a trend for improvement in isometric muscle strength (i.e. hip extension, hip abductor) were also observed at either 6, 12 or 18 weeks. Small effect sizes were detected in time spent sitting/lying, standing and MCS. A single-centre study rarely provides sufficient evidence to guide clinical and research practice; nevertheless, the results of this study do add to the findings of other previous CP exercise studies. It is clear that further, appropriately powered research is warranted.

CHAPTER 8 FINAL DISCUSSION

8.1 Introduction

The primary aim of this thesis was to investigate the effects of an 18-week exercise programme on the physical function, gait quality, habitual physical activity, quality of life and self-esteem of adolescents and young adults with CP through a single-blind randomised controlled trial (Study 3, Chapter 7). The study's secondary objectives were to evaluate (a) the methodological quality and strength of evidence of the published articles that reported the psychometric properties of gait quality and walking performance in adolescents and young adults with CP using a standardised quality checklist (COSMIN) (Study 1, Chapter 4), and (b) the reliability of physical function, habitual physical activity, quality of life and self-esteem outcome measures in adolescents and young adults with CP, using a test-retest study design (Study 2, Chapter 5).

The key findings from the preceding chapters are summarised in Table 8.1. In each of the preceding four chapters the empirical studies have been discussed independently of one another. The purpose of this chapter is therefore to synthesise and integrate these chapter-specific findings and to discuss these in the broader context of the existing literature. The implications for clinical and research contexts are discussed along with an identification of the limitations of this study and feasibility issues that will inform future work.

Table 8.1 Key findings from the preceding four chapters

Chapter	Key findings
Chapter 4: Psychometric properties of measures of gait quality and walking performance in young people with cerebral palsy: A systematic review	• Fourteen OMs for measuring gait quality and walking performance were identified. Strong and moderate levels of evidence were found for the reliability of FMS and validity of GPS, respectively. • No evidence was found for responsiveness and measurement error for any OMs.
Chapter 5: Reliability of physical function, habitual physical activity, quality of life and self-esteem outcome measures in young people with cerebral palsy and age-matched controls	• The ICC values for participants with CP were: TUG (0.95, 95% CI 0.77–0.99), 10m SRT (0.86, 95% CI 0.44–0.97), isometric hip extensor strength (0.89, 95% CI 0.56–0.98) and HPA (sedentary behaviour, standing, stepping, step counts, sit to stand) measured by an activity monitor (ActivPAL^{TM}) (0.76–0.86, 95% CI 0.02–0.98) when assessed 6 weeks apart in adolescents and young adults with CP with GMFCS level I–III. Moderate reliability was found for the SF-12 physical component score (ICC=0.74, 95% CI -0.03–0.96). The ICC for the SF-12 mental component score was 0.45 (95% CI -0.45–0.90) and for the RSES was 0.13 (95% CI -0.69–0.81), indicating poor reliability. • The ICC values for the healthy controls were: TUG (0.80, 95% CI 0.48–0.93), 10m SRT (0.92, 95% CI 0.77–0.97) and RSES (0.96, 95% CI 0.89–0.99). Moderate reliability was found for hip abductor strength (0.57, 95% CI 0.08–0.84), PCS (0.53, 95% CI 0.03–0.82) and MCS (0.57, 95% CI 0.08–0.84). Poor reliability was found for hip extensor strength (0.36, 95% CI -0.19–0.74), knee extensor strength (0.16, 95% CI -0.39–0.62) and HPA (0.24-0.50, 95%CI -0.39–0.84).

Chapter	Key findings
	• The MDC_{95} and MDC% for OMs of physical function, habitual physical activity, quality of life and self-esteem in adolescents and young adults with CP and age-matched controls were established.
Chapter 6: Physical function, habitual physical activity, quality of life and self-esteem in young people with cerebral palsy and age-matched controls: A cohort study	• This study confirms that the young people with CP in our study had significantly lower physical function, lower habitual physical activity and reported a lower physical component of QoL than their age-matched peers. However, the mental component score and self-esteem were not significantly different between the CP group and healthy controls.
Chapter 7: Effects of an 18-week pragmatic community exercise programme on physical function, gait quality, habitual physical activity, quality of life and self-esteem of young people with CP: a single-blind RCT	• No significant time x group interaction for any outcome measures at either 6 weeks (for physical function) or 12 weeks (for HPA, SF-12, RSES, COPM, FAQ); small effect size found in the COPM (*d*=0.54) at 12 weeks. • Considering the experimental group only: statistically significant improvement in the COPM at 12 weeks (p=0.02, *d*=0.81) and trends indicating increased isometric strength of hip extensor (*d*=0.67) and abductor muscle (*d*=0.54), decreased time spent sitting/lying *(d*=0.36), increased time spent standing (*d*=0.36) and increased SF-12 MCS (*d*=0.34) at 12 weeks. No statistically significant changes in gait quality (p=0.62, *d*=0.29), step count (p=0.5, *d*=0.28), sit to stand (p=0.9, d=0.07), SF-12 PCS (p=0.8, *d*=0.28) and self-esteem (p=0.2, *d*=0.09) over time.

FMS: Functional Mobility Scale; GPS; Gait Profile Score; 3DGA: Three-Dimensional Gait Analysis; OMs: Outcome Measures; ICC: Intraclass Correlation Coefficient; TUG: Timed Up And Go Test; 10m SRT: 10-metre Shuttle Run Test; HPA: Habitual Physical Activity; GMFCS: Gross Motor Function Classification Score; MDC: Minimal Detectable Change; COPM: Canadian Occupational Performance Measure; d: Cohen's d effect size; GMFM: Gross Motor Function Measure; QoL: Quality of Life; PCS: Physical Component Score; MCS: Mental Component Score; SF-12: Short Form 12

8.2 Current psychometric evidence supporting outcome measures of gait and walking performance in young people with CP

Clinicians and researchers should be looking for evidence from high-quality psychometric studies to reassure them of the validity, reliability and responsiveness of the outcome measure(s) they decide to use. However, Study 1 (Chapter 4) showed that there is a lack of such high-quality studies reporting the psychometric properties of gait quality and walking performance outcome measures used in young people with CP. As a result, the systematic review in Chapter 4 concluded that only the reliability (consistency) of the Functional Mobility Scale (FMS) has a 'strong' level of evidence and the construct validity of the Gait Profile Score (GPS) derived from the 3DGA has a 'moderate' level of evidence. The rating of the methodological quality in many studies was reduced from 'excellent' to 'good'/'fair', often due to poor choices for the statistical analysis. The results from Study 1 also demonstrated that the evidence for 14 OMs with regard to responsiveness and measurement error, including for FMS and GPS, remains unknown.

Responsiveness was reported in only three of the studies included in the systematic review (Chapter 4), and the methodological quality of all three was rated as 'poor' for the reason of not including a comparator outcome measure. Moreover, none of these three studies reported Minimal Clinically Important Change (MCID) values. The limited choice of outcome measures with robust psychometric properties, especially pertaining to responsiveness, is of concern as any ability to assess changes in gait quality and walking performance in patients following an exercise intervention requires outcome measures that are responsive to change. In addition, it was concluded that there is a lack of studies reporting measurement error (e.g. MDC).

Concern about the lack of or unknown evidence, especially with regard to responsiveness, was identified not just in the current review but also in previous systematic reviews with CP populations (Ketalaar et al. 1998, Harvey et al. 2008, Carlon et al. 2010). In their systematic review of functional motor abilities outcome measures, Ketalaar et al. (1998) suggested that there is an urgent need to explore measures that can evaluate change in functional abilities in children with CP. Harvey et al. (2008) suggested that more studies on the responsiveness of activity limitation outcome measures are needed for children with CP. Another systematic review by Carlon et al. (2010) on the quality-of-life measures for school-aged children with CP also recommended more work on data for sensitivity to change. In addition, Carlon et al. (2010) also commented on the lack of available data on measurement error despite the fact that the methodological quality of studies assessing validity and reliability with regard to consistency is improving.

The clinical significance of a difference in a certain measure after treatment is represented by the MDC and MCID (de Vet et al. 2006). MDC is defined as the smallest change beyond measurement error, and any change higher than the MDC can thus be regarded as a true difference (de Vet al. 2006). MCID is defined as the smallest change in an OM that is perceived to be effective by the patient or clinician (de Vet et al. 2006, Guyatt et al. 2002, Jaeschke et al. 1989). The MDC is derived from test-retest reliability studies while the MCID is derived from the results of a longitudinal study where outcomes are expected to change over time as a result of a certain intervention. Therefore, the values for MDC and MCID provide clinicians with the information they need for effective clinical decision-making when interpreting score changes following a certain intervention. While this thesis presents the MDC values for all of the OMs included in Study 2, the values for the MCIDs were not derived. There were two reasons for this. Firstly, it is not recommended to combine a study on the responsiveness of outcome measures with an intervention study, as it is not

known whether the intervention actually brings about a change in outcome. Secondly, there are methodological issues regarding the way in which MCID is derived. The anchor approach has been recommended to calculate MCID (Copay et al. 2007, Haley and Fragala-Pinkham 2006b) compared to the distribution approach. However, as there is a lack of a gold standard in the area of health care and rehabilitation, the anchor is often a patient or clinician rating scale such as the Global Rating Scale (Kamper 2009), and the evidence regarding the validity of such a rating scales is currently conflicting (Kamper 2009).

8.3 Reporting standards of clinical studies

Several standards are currently available to help with the reporting of research and clinical studies, including 1) Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) (Moher et al. 2016, Moher et al. 2015), 2) Consolidated Standards for Reporting Trials (CONSORT) (Schulz et al. 2015), and 3) Revised Standards for Quality Improvement Reporting Excellence (SQUIRE 2.0). More details on these and other guidelines can be found in the EQUATOR (Enhancing the QUAlity and Transparency Of health Research) website (http://www.equator-network.org). These standardised checklists guide the author on the reporting or designing (i.e. methodological aspects) of intervention studies but not of psychometric studies. The COSMIN checklist, as detailed in Chapter 4, is a specific checklist concerned with the methodological quality of psychometric studies. It incorporates a multi-level grading system for each measurement property, thus giving detailed information on the quality of outcome measures as well as the appropriate usage of statistical analysis for specific measurement properties. The COSMIN checklist is therefore also aimed at informing clinicians and researchers with regard to the reporting and design of psychometric studies.

The COSMIN checklist was originally developed to rate the methodological quality of health-related quality of life surveys. However, in the systematic review in this thesis and many other systematic reviews, the methodological quality of non-questionnaire-based outcome measures has been rated using the COSMIN checklist, and this is regarded as a limitation by some researchers. Although another standard assessment to rate studies on the measurement properties of outcome measures has been published by Brink and Louw (2012), this was not developed for use in systematic reviews. The COSMIN checklist was developed for non-questionnaire-based outcome measures; as such, there were items in the checklists which were not applicable to the current review, such as missing items. Internal consistency and content validity were not rated/not applicable. This was discussed in Chapter 4. Apart from that, all other boxes (measurement properties) and items on the COSMIN checklist are applicable for all types of outcome measures.

8.4 Contribution of this thesis to psychometric evidence for outcomes of physical function and physical activity, QoL and self-esteem in young people with CP

To address some of the gaps in the evidence regarding the lack of data on measurement error in commonly used outcome measures identified in the systematic review (Chapter 4) and previous systematic reviews, Study 2 (Chapter 5) was performed. In this study the reliability (both consistency and agreement/measurement error) of outcomes of physical function (TUG, 10m SRT, isometric muscle strength, habitual physical activity (ActivPAL^{TM}), QoL and self-esteem outcome measures in adolescents and young adults with CP were determined through a test-retest design with a 6-week interval. It was found that the ICC of the TUG was 0.95 (95% CI 0.77–0.99), that of the 10m SRT was 0.86 (95% CI 0.44–0.97) and there was an ICC of 0.89 (95%CI 0.56–0.98) for isometric hip extensors. The ICCs for the isometric hip abductors and habitual

physical activity were between 0.75 and 0.86 (95% CI 0.02–0.98), indicating good reliability ('consistency'). The physical component scores of the SF 12 were found to have moderate reliability (ICC=0.74, 95% CI -0.03–0.96). These findings are supported by other studies, which found good reliability of the TUG (Dhote et al. 2012, Williams et al. 2012, Gan et al. 2008), 10m SRT (Verschuren et al. 2006, Verschuren et al. 2011), isometric hip extensor strength (Seniorou et al. 2002) and ActivPAL^{TM} (Bania 2004) in the CP population. However, the majority of these previous studies used an interval of less than 6 weeks, with the majority having a one-week gap between tests. The 6-week interval in our study was selected as this interval is often selected in exercise intervention studies since it is often believed that around 6 weeks is sufficient for an exercise intervention to induce changes in physical fitness (Taylor et al. 2013).

Therefore, our findings contribute to the evidence in the literature regarding the reliability (consistency) of the physical function, isometric muscle strength and ActivPAL^{TM} in young people with CP. However, the ICCs of the mental component score of the SF-12 and self-esteem measured by the RSES were 0.45 (95%CI -0.45–0.90) and 0.13 (95%CI -0.69–0.81) respectively, thereby indicating poor reliability. Nevertheless, the current study provides the MDC values for SF-12 and RSES, and this could help clinicians or researchers to interpret the changes (i.e. a true change or measurement error). No previous studies have examined the reliability of SF-12 and RSES in adolescents and young adults with CP; hence, future studies are needed to confirm our results.

In addition to the consistency aspect of reliability, the current study also reported the MDC_{95} values (i.e. 'agreement aspect of reliability') for all of the outcome measures included in Study 2, Chapter 5. The MDC_{95} values found in Study 2 in relation to the changes found in the exercise study (Study 3, Chapter 7) are shown in Table 8.2. None of the average changes found in Study 3 were beyond the MDC_{95} values reported in Study 2. This may indicate that the changes found

in Study 3 were possibly due to measurement error and do not reflect a true change as a result of the exercise programme. On an individual level, however, a few of the participants exceeded the MDC_{95} values in different outcome measures (Table 8.2).

With regard to the step counts, the MDC_{95} reported in Study 2 was 2667 steps/day, and this value is quite high in comparison to that for other populations using the same activity monitor, i.e. ActivPAL^{TM}. Van der Linden et al. (2013) found the MDC_{95} for step counts in people with multiple sclerosis was 1741 steps/day. So far, no study has reported the MDC of habitual physical activity pattern using ActivPAL^{TM} in the CP population. A possible explanation for this high measurement error may be that habitual physical activity is complex and is related to psychological, social or environmental factors and physical factors (Palisano et al. 2011, Bania et al. 2011, Anderson-Bill et al. 2011, Conchar et al. 2016), meaning that daily step count, even when averaged over a period of 4 to 7 days, can vary considerably from week to week.

In summary, Study 1 (Chapter 4) identified a lack of studies reporting the MDC of measurements used in the population with CP, while Study 2 was the first to report the absolute and relative MDC values of measures of physical function, habitual physical activity patterns, quality of life and self-esteem in young people with CP. The results from Study 2 can be used as a reference for the measurement error of measures of physical function (i.e. TUG, 10m SRT, isometric muscle strength) and habitual physical activity patterns (i.e. activity monitor ActivPAL^{TM}) to help clinicians and researchers determine the 'true' change between two assessments.

Table 8.2 The minimal detectable change values reported in Study 2 and Study 3

Outcome measures	MDC_{95} values in Study 2, Chapter 5 with 6-week interval	Mean (SD) changes found in Study 3, Chapter 7 (experimental group)	Number of participants in the exercise group exceeding the MDC
TUG(s)	6.88	2.3[a]	1/9
10m SRT (nr of shuttles)	3.6	0.3[a]	1/9
Hip extensor (Nm/Kg)	0.6	0.05[a]	1/9
Hip abductor (Nm/Kg)	0.44	0.09[a]	0/9
Knee extensor (Nm/Kg)	0.74	0.27[a]	1/9
GMFM 66 Dimension D&E	3.62	2[a]	2/9
GPS (°)¥	5.6	0.13[b]	0/9
Sitting/lying* (hours/day)	1.8	0.6[b]	1/5
Steps/day*	2667	784[b]	1/5
Sit to stand* (number/day)	13.6	0[b]	2/6
COPM (Performance 0-10)	1	1.5[b]	2/7
FAQ (0-25)	3	1[b]	1/7
PCS (34.75-56.71)µ	13.21	1.3[b]	0/4
MCS (15.37-62.39)µ	22.26	5.5[b]	¼
RSES (0-30)	7.56	0.8[b]	0/4

*MDC, Minimal Detectable Change; TUG, Time Up and Go test; 10m SRT, 10-metre shuttle run test; GMFM, Gross Motor Function Measure; GPS, Gait Profile Score; COPM, Canadian Occupational Performance Measure; FAQ, Gillette Functional Assessment Questionnaire; PCS, Physical Component Score; MCS, Mental Component Score; RSES, Rosenberg Self-Esteem Scale;¥derived from kinematics data from three-dimensional gait (3DGA) analysis; *derived from activity monitor known as ActivPAL™ ; µderived from Short Form 12 version 2 (SF-12); [a] 6-week interval; [b] 12-week interval*

8.5 Exercise programme for adolescents and young adults with CP

Study 3 (Chapter 7) was conducted in response to the paucity of research with scientifically robust designs that investigated exercise interventions in adolescents and young adults with CP. This is unfortunate since adolescence and early adulthood seems to be a crucial time at which to implement this type of management strategy. This stage of life appears to mark the beginning of functional decline and greater social isolation for people with CP (Ng et al. 2003, Stevenson et al. 1997, Jahnsen et al. 2004, Liptak 2008). Additionally, it is during this time that people with CP must begin the transition from child health care to adult health care. Introducing adolescents and young adults with CP to an exercise programme would perhaps provide them with a means of self-managing their health and well-being throughout their adult life, when the provision of physiotherapy is rare and often absent. Therefore, the main aim of this study was to evaluate the feasibility and effectiveness of an 18-week exercise programme on physical function, gait quality, habitual physical activity, quality of life and self-esteem in adolescents and young adults with CP.

Four hundred and thirty ambulant young people with CP aged 16 to 25 years were invited to participate in this study. However, only 19 adolescents and young adults with CP were randomly allocated to either the exercise (n=10) or control groups (n=9) and assessed at baseline. At the 6-week assessment, nine participants in the exercise group and seven participants in the control group were available for analysis; by the 12-week assessment this had fallen to seven and five participants, and at 18 weeks the numbers were seven and one for the exercise and control groups, respectively. Due to the recruitment and retention of the participants in the study being lower than anticipated, analysis of the RCT was performed for the baseline and 6-week assessments. The results of the 12- and 18-week assessments were explored using a within-group analysis for the

experimental group only. The results showed no statistically significant effects for the group x time interaction in the analysis of the RCT at 6 weeks. Within-group analysis of the experimental group showed a statistically significant improvement for only the self-reported function (COPM) (p=0.02) at 12 weeks compared to the baseline.

8.5.1 Physical function, physical activity, QoL and self-esteem in young people with CP compared to age-matched healthy peers

In Chapter 6, the baseline of all OMs between the CP group (both exercise and control groups) and healthy controls was explored. Statistical analysis (Mann-Whitney test) showed that the CP group was significantly weaker, walked slower, had a lower level of aerobic fitness and a lower level of habitual physical activity than their healthy age-matched peers.

In order to put any changes due to the exercise programme into perspective, Figures 8.1–8.3 show the outcome measures of the exercise group (baseline and 6- & 12-week assessments, post exercise) and those of the healthy age-matched peers. Not surprisingly, regarding the lack of exercise-induced effects, none of the physical function measures of the exercise group post exercise reached the values of the healthy group. Knee extensor strength at week 6 in the exercise group was the closest to the healthy group compared to other measures of physical function. Similarly, none of the post-exercise programme measures of habitual physical activity in the exercise group reached those of the healthy controls.

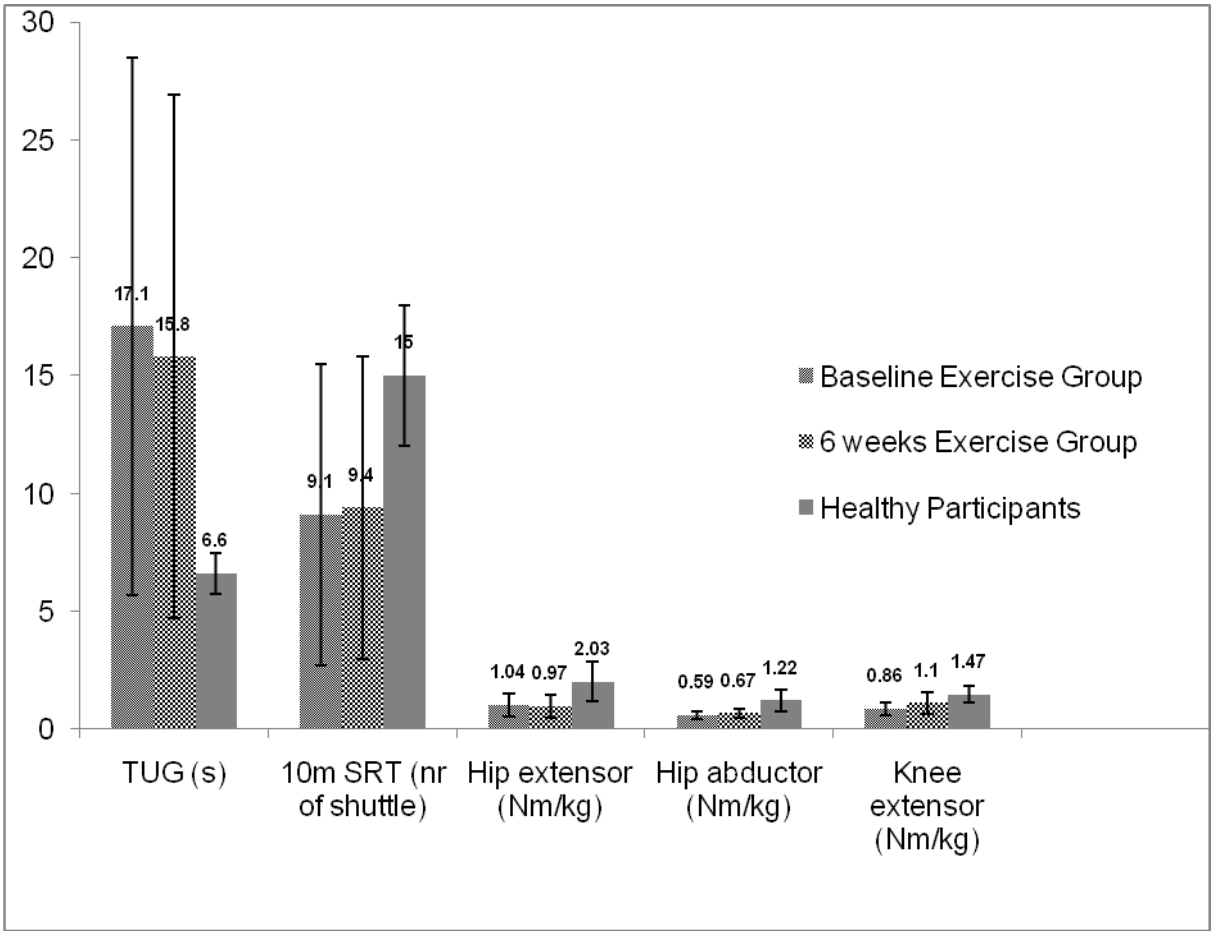


Figure 8.1 Comparison of the physical function measures between the exercise group (pre and post exercise) and healthy participants

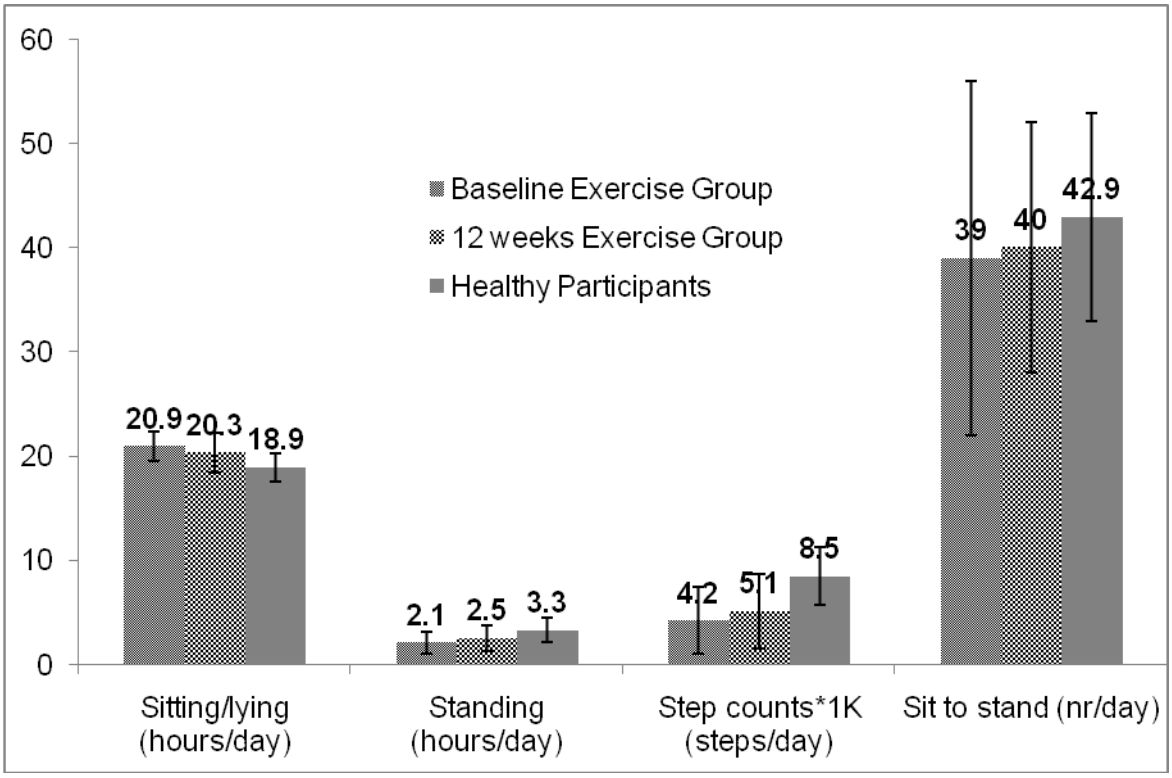


Figure 8.2 Comparison of the habitual physical activity pattern measures between the exercise group (pre and post exercise) and healthy participants

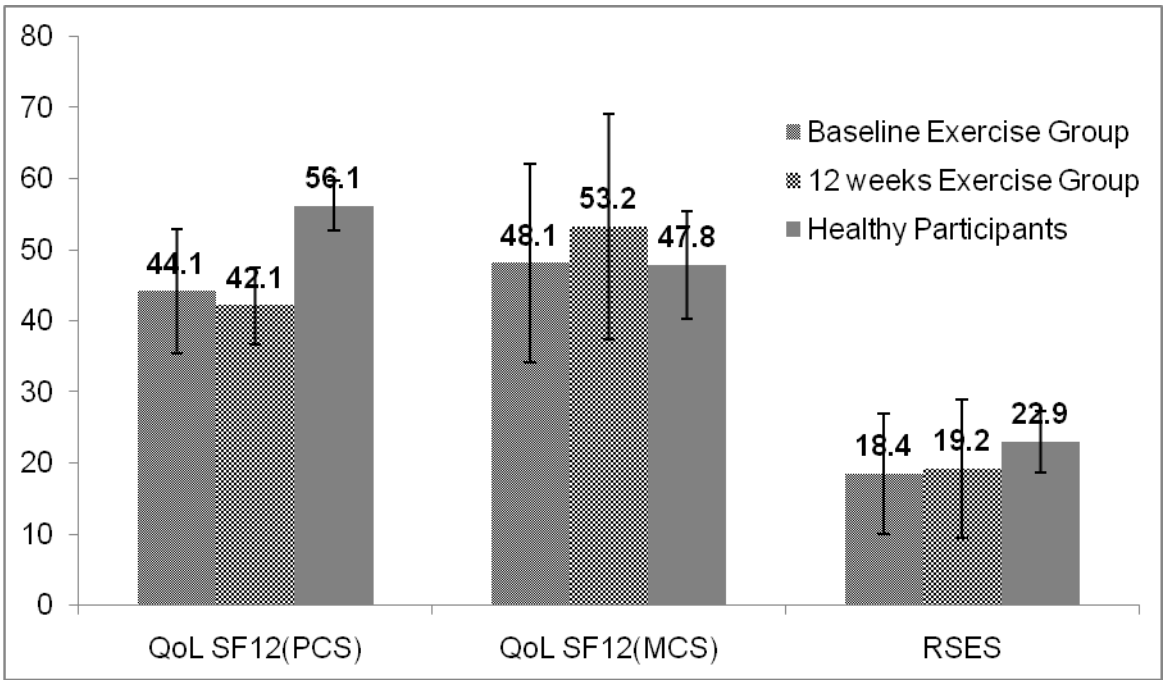


Figure 8.3 Comparison of the QoL and self-esteem measures between the exercise group (pre and post exercise) and healthy participants

8.5.2 Habitual Physical Activity

Habitual physical activity is an important outcome for any study seeking to explore the effects of an exercise programme. Increasing the levels of physical activity in people with CP is imperative as it may potentially reduce the risk of secondary health problems such as cardiovascular disease, obesity and osteoporosis (Kokkinos 2012). In addition, increasing physical activity levels may also decrease some of the problems commonly associated with CP such as muscle weakness, stiffness and decreased mobility (Thompson et al. 2011, Kerr et al. 2011). The evidence suggests that participating in an exercise programme can increase the daily physical activity of people with CP (Bania et al. 2011, Van den Berg-Emons et al. 1998).

Habitual physical activity in this study was captured using an activity monitor known as ActivPAL^{TM}. From this monitor, the time that the participants spent 1) sitting/lying and 2) 'stepping' (standing and walking), plus 3) number of step counts, and 4) the number of standing-to-sitting transitions per day between four and seven days were derived. Study 3 revealed that there was no statistically significant interaction between group and time in any of the measures of habitual physical activity, and this result is consistent with that obtained in a study by Bania et al. (2016) on participants with CP aged between 14 and 22 years. A possible explanation for this lack of an increase in habitual physical activity in the intervention group is the phenomenon of resting after a bout of exercise, as described by (Rowland 1998). Participants may have felt they had worked hard during their weekly training sessions and thus on their non-gym days they felt they deserved or needed more rest. Informal anecdotal feedback from the carer of one of the participants indicated that her daughter spent most of her time sitting on the couch watching television (on non-exercise days) because she felt too tired after spending time at the gymnasium 2 to 3 times per week.

Another issue associated with using habitual physical activity as an outcome measure in exercise studies is the high day-to-day or even week-to-week variability in this measure, as was found in Study 2, which confirmed the results of previous studies (Bania et al. 2016, van der Linden et al. 2014). The high measurement error was also shown by an MDC of 2667 steps/day in Study 2 and 1741 steps/day in a study of people with multiple sclerosis (van der Linden et al. 2014).

Further, addressing lower limb strength and aerobic capacity alone, without addressing any psychological, social or environmental factors, may not have been sufficient to elevate the habitual physical activity measures and reduce sedentary time in people with CP. Combining the exercise training and patient education and/or counselling on physical activity participation may be more

effective in changing an individual's physical activity behaviour (van der Ploeg et al. 2006, Slaman et al. 2010, Van der Ploeg, Hidde et al. 2007). A recent multi-centre RCT with 57 participants with CP aged 16–25 years revealed significant improvement in self-reported physical activity measured using the Physical Activity Scale for Individuals with Physical Disabilities following a six-month lifestyle intervention consisting of physical fitness training combined with counselling sessions that focused on physical behaviour and sports participation (Slaman et al. 2015). However, this effect was not maintained in the long-term follow-up (12 months), and no significant improvement was found in the objective habitual physical activity measured using an accelerometer. Whilst a combination of exercise training and patient education seems promising with regard to improving the level of physical activity among the CP population, further studies are clearly needed to confirm this.

8.5.3 Sustainability/maintenance of exercise-induced effects and exercise behaviour

The short-term effects of an exercise programme with regard to physical activity have been explored in both this thesis and previous CP exercise studies (Slaman et al. 2015, Bania et al. 2016). However, the long-term effects of such programmes remain unknown. One of the long-term aims of an exercise programme is the achievement of a behavioural change towards a more active lifestyle. The aim of the design of the current study, with a reduction in the prescribed number of weekly exercise sessions over the three 6-week blocks, was to induce exercise behaviour that would be maintained beyond the duration of the 18-week programme.

Although a long-term (>12 months), formal assessment of any possible exercise-induced effects was beyond the scope of this thesis (i.e. it was not possible within the 3-year PhD programme), an informal follow-up (i.e. telephone

and email) at approximately 2 months following the completion of the 18-week programme was carried out. Former participants were asked whether they were still engaged in any exercise or physical activity after completion of the programme. Due to problems with reaching the participants via telephone and email, only two participants out of four (from the 2015–2016 batch) provided feedback. One participant reported cycling to college 3x/week and doing sports activity, i.e. indoor/outdoor climbing, while another participant replied that she only took part in physical education at school. The current study did not provide an objective measure of habitual physical activity patterns to identify the long-term effects of the programme. Direct observation through activity monitoring is probably the most appropriate and practical measure of assessing habitual physical activity patterns (Sirard and Pate 2001) in order to capture the long-term effects of an exercise programme in people with CP and should therefore be considered for future research.

8.5.4 Feasibility issues

8.5.4.1 Recruitment and participant retention

A total of 430 ambulant young people with CP aged 16 to 25 were invited to participate in this study, resulting in a total of 28 individuals initially expressing an interest in participating. However, only 19 adolescents and young adults with CP were randomly allocated to either the exercise (n=10) or control groups (n=9) and assessed at baseline. At the 12-week assessment, seven participants in the exercise group and five participants in the control group were available for analysis.

This very low recruitment rate is not unique to either the current study or the wider CP population and has also been reported in previous exercise studies conducted with this age group in the general population or other patient groups (Rooks et al. 2006, Fairey et al. 2005, Heinrichs et al. 2005).

There were probably two issues with the low recruitment. Firstly, there was the issue of the participants' willingness to take part in a research study which required four assessments, each of 2 to 3 hours in duration, over a period of 18 weeks. Although no further survey was carried out enquiring as to the reasons for the low recruitment, one participant who withdrew prior to the baseline assessment taking place informed that it was due to his limitations in terms of the session scheduling and location (i.e. for assessments) rather than an inability to participate in the exercise programme. The use of simple, quick and easy measurement tools for walking performance may thus have helped to partially overcome this barrier to taking part in exercise studies (Himuro et al. 2015). The measurement tools recommended by Himuro et al. (2015) are TUG, the one-minute walk test, FAQ, ABILOCO-Kids and FMS; however, the measurement error and responsiveness are yet to be confirmed for these measures.

The second issue was the participants' willingness to take part in a gym-based exercise programme. To promote an active lifestyle in this population, it is crucial to understand the perceived barriers to physical activity in people with CP. Some of the barriers to participation in physical activity in children and adolescents with CP have been reported as being challenges with transportation to the sports club (Verschuren et al. 2012, Lawlor et al. 2006), the attitudes of strangers and staff in public places, the presence of stairs (Lawlor et al. 2006) and financial constraints (Verschuren et al. 2012). Other studies with young and older adults aged 18 to 40 years with physical disabilities reported perceived barriers to participate in physical activity such as time constraints, lack of energy, fear of injuries and too much effort required (Buffart et al. 2009, Rimmer et al. 2004).

8.5.4.2 Dropout in the exercise group

Two participants dropped out of the programme, one due to depression and one due to chronic fatigue syndrome. It was noted at the baseline assessment that the latter participant had suffered from fatigue over the course of past few years. Notes in the exercise logbook also showed that the participant felt tired from the second week of the programme onwards. The participant had been exercising prior to the study, comprising mainly swimming at the leisure centre. As the exercise programme in the study prescribed an aerobic, land-based form of exercise, the participant had to travel a few miles further to use a Motomed exercise bike, as she was not able to use the usual aerobic equipment (recumbent bike, rower, treadmill) at her local leisure centre. It is possible that this change to a less convenient venue was more demanding than her usual exercise routine.

Optimal participant retention is an important factor to consider in the feasibility of an RCT. Excessive attrition can limit the meaningful interpretation of results. In this study, two of the participants in the exercise group and three of the participants in the control group withdrew from the programme (at different time points, refer to Figure 7.3), resulting in an attrition rate of 29%. Dropout in previous CP exercise studies ranged from 4% to 35% (McNee et al. 2009, Taylor et al. 2013, Bania et al. 2014, Slaman et al. 2015). Its attrition of greater than 20% indicates that the results of this study have a high risk of bias and should therefore be treated with caution (Dumville et al. 2006).

8.5.4.3 Adherence to the exercise programme

Adherence to a thrice-weekly followed by a twice-weekly exercise programme was confirmed by the excellent compliance with the thrice-weekly sessions during the first six weeks (86%) and in the second block of two weekly sessions (90.6%). The majority of the participants (n=7 out of 9) continued with exercise

sessions in the last six weeks (12–18 weeks), where there was no prescribed number of sessions. However, gymnasium attendance fell considerably, with some of the participants failing to attend the gym at all in those weeks.

The reduced gymnasium attendance in weeks 12–18 could be partly due to the lack of input from the physiotherapist. Evidence shows that people with a disability feel safer and more confident when exercising with the involvement of a physiotherapist (Lennon et al. 2013, Simpson et al. 2011). In addition, one participant commented, *'…..GM (physiotherapist) was a great support'* in the feedback questionnaire, thus indicating that the input of a physiotherapist is an important factor influencing the success (adherence, fidelity) of the exercise programme. Future studies may perhaps wish to consider the inclusion of contact with a physiotherapist for their participants at least once during weeks 12 to 18.

8.5.4.4 Perception of the exercise programme

Despite the lack of statistically significant effects both between groups (in the 6-week RCT) and within the experimental group at 6, 12 and 18 weeks in all OMs except the COPM, the exercise programme feedback questionnaire showed a positive response with regard to the programme, as shown in Table 7.5 (Chapter 7). In particular, one participant commented on the feedback questionnaire as follows: '*This programme was a good idea. I was feeling motivated, to keep myself fit, happier.'* Durstine et al. (2000) suggested that enjoyment is an important element when seeking to implement an exercise programme. Previous CP exercise studies also found significant increases in their participants' self-perception (Verschuren et al. 2007, Unger et al. 2006, Dodd et al. 2004), as well as them expressing enjoyment and feeling more confident by participating in the exercise programme (Mc Burney et al. 2003, Allen et al. 2004, Morton et al. 2005). Further, enjoyment of the exercise programme is an important factor for

young adults in terms of them deciding whether or not to continue with the exercise programme on a regular basis in the future. According to previous studies (Allenet al. 2004, McBurney et al. 2003, Lee et al. 2008, Unger et al. 2006), future exercise studies should consider incorporating a qualitative analysis (i.e. interview or focus group) when determining the effectiveness of such a programme and the factors determining effectiveness.

8.5.4.5 Selection of outcome measures

The selection of the range of outcome measures used in this study was justified through their psychometric properties (validity and reliability), their feasibility (practicality) and the fact that the OMs represented all three of the domains described in the ICF framework (as discussed in Chapter 2). It is possible, however, that other measures may have been more appropriate to reflect any improvements induced by the exercise programme and that such measures should be considered in future research. For example, to assess quality of life, a disease-specific questionnaire (i.e. TACQOL) may be more effective for assessing those areas of life that may be most affected by the specific disease or condition. However, generic questionnaires give a broad assessment of health status and allow for the comparison of health-related QoL between groups of patients with different conditions. Ideally, it is recommended to combine both generic and disease-specific tools. Regarding the low reliability of SF-12 and RSES, this is indeed a limitation of the thesis, as the reliability study was conducted after the first batch of the intervention study. Using the myometer to measure isometric muscle strength in individuals with CP, the effects of their spasticity probably affected the participants' strength measures, although spasticity has been found to be independent of strength (Ross and Engsberg 2002). Despite this, strength testing has demonstrated good reliability (consistency) with a high measurement error in this population, as shown in Study 2. Instead of TUG, the 6-minute or 2-minute walk tests are probably better

tools to use to evaluate walking, since in the list of activities in the COPM that participants wanted to improve, seven of the sixteen participants indicated that they would like to improve their walking distance. So while the studies in this thesis did not intend to present an exhaustive list of possible measures, they do provide examples of how clinical measures that are appropriate to the research setting (i.e. laboratory) relevant to adolescents and young adults with CP can be considered in the context of the ICF model.

As discussed in previous sections, such alternative measures should have been reliable and responsive to change. In particular, it is recommend that the responsiveness and in particular the MCID of these outcome measures are established. Table 8.3 summarises the limitations identified in the current study and some potential solutions for use in future research.

Table 8.3 Main limitations identified throughout the thesis

Limitations	Recommendations
The sample size in the exercise study was small	• Multi-centre trial • Decrease assessment burden to improve recruitment. Reduce barriers and reinforce facilitators to exercise.
Responsiveness (MCID) was not explored	• A responsiveness study with appropriate methodology design and statistical analysis should be conducted to inform clinicians of the clinically important change.
No measurement of physical activity in medium-/long-term follow-up	• Follow up at 24 weeks & 12 months with ActivPAL^{TM} to determine habitual physical activity.
Exercise programme solely focused on improving physical fitness and habitual physical activity	• Apart from the exercise programme, future studies could perhaps incorporate counselling/patient education on physical activity or participation in sports on an individual basis.

MCID: minimal clinically important change

8.6 Conclusion

The need to use outcome measures that have robust psychometric properties to evaluate the effectiveness of interventions such as strength and aerobic training is well recognised. However, only two of the outcome measures currently being used to measure gait quality and walking performance in young people with CP were found have strong to moderate evidence in terms of their reliability and validity, with very limited information and evidence available on their responsiveness and measurement error (Chapter 4).

The results from the reliability study (Chapter 5) add to the evidence base for the psychometric properties (i.e. consistency and agreement) for the outcome measures used to measure physical function and habitual physical activity; however, more studies are needed to investigate these psychometric properties for the different QoL and self-esteem outcome measures used for young people with CP.

The exercise study (Chapter 7) explored the feasibility of an 18-week exercise programme in young people with CP. In addition, the effect on physical function, gait quality, habitual physical activity, QoL and self-esteem following the exercise programme was explored with inferential analysis, and the effect sizes for each outcome measure were reported. Further work is still required to confirm these findings in a larger sample. Other limitations, as well as recommendations, have been presented for both clinicians and researchers for future direction, as presented in Table 8.3.

In conclusion, the series of studies in this thesis has served to answer some of the questions related to the level of evidence for gait quality and walking performance outcome measures, the psychometric properties (i.e. reliability) of the OMs used in exercise studies and the effects as well as the feasibility of an 18-week exercise programme on adolescents and young people with CP. These studies have also generated directions for further exploration in

CP research; in particular, investigations on the responsiveness (MCID) and the integration of patient education in the exercise programme, in addition to the need to follow up (i.e. over the medium and long terms) in order to gain information on the sustainability of increases in the physical activity levels of people with CP.

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Appendices

Appendix 1 GMFM scoring instruction

Test should be performed **without** shoes on
Participant gets a max of 3 attempts for each item and best performance is scored – test trials can be used

STANDING

52.On the Floor: Pulls to stand
Starting position: any on floor other than standing
Ending: can be leaning on bench

0 = does not initiate standing
1 = initiates pulling to stand
2 = partially pulls to stand
3 = pulls to stand

53.Standing: Maintains, arms free, 3 sec
0 = does not maintain standing,
1 = maintains, 2 hands holding on, 3 sec
2 = maintains, 1 hand holding on, 3 sec
3 = maintains, arms free, 3 sec

54/55.Standing: Holding on to large bench with one hand, lifts right/left foot, 3 seconds
0 = does not initiate lifting right/left foot
1 = holding bench 2 hands, lifts right/left foot, <3 s
2 = holding bench 2 hands, lifts right/left foot, 3 s
3 = holding bench 1 hand, lifts right/left foot, 3 sec

For 3 must **begin** holding with only one hand

56.Standing: Maintains, arms free, 20 seconds
0 = does not maintain standing, arms free
1 = maintains, arms free, <3 sec
2 = maintains, arms free, 3-19 sec
3 = maintains, arms free, 20 sec

57/58.Standing: Lifts left/right foot, arms free, 10 seconds
0 = does not lift left foot, arms free
1 = lifts left foot, arms free, <3 sec
2 = lifts left foot, arms free, 3-9 sec
3 = lifts left foot, arms free, 10 sec

59.Sitting on small bench: Attains standing without using arms
Starting: feet flat on floor, knees approx. 90°

0 = does not initiate standing
1 = initiates standing
2 = attains standing using arms
3 = attains standing without using arms (on body or bench)

60/61.high kneeling: attains standing through half kneeling on right/left knee, no arms
Starting: high kneeling – weight bearing on knees, buttocks clear of lower legs

0 = does not initiate standing
1 = initiates standing
2 = attains standing using arms (start from high kneel, but not necessarily through half knee)
3 = attains standing through half kneeling on right/left knee, without arms (on body or ground)

62.Standing: Lowers to sitting on floor with control, arms free
Starting : must start standing free, if hold on to lower must only be once movement is initiated

0 = does not lower
1 = lowers to sit on floor, but 'crashes' down
2 = lowers to sit on floor with control, uses arms or holds on
3 = lowers to sitting on floor with control, arms free

63.Standing: Attains squat, arms free
Starting : must start standing free, if hold on to lower must only be once movement is initiated

0 = does not initiate squat
1 = initiates squat
2 = attains squat, using arms or holding on
3 = attains squat, arms free

Squat = crouching close to ground with hips and knees flexed at least 90°

64.Standing: Attains squat, arms free
0 = does not initiate picking up object
1 = initiates picking up object
2 = picks up object, using arms or holding on (includes leaning on floor)
3 = picks up object, arms free, returns to standing

WALKING RUNNING & JUMPING

65/66. Standing two hands on bench: Cruises five steps to the right/left
Starting: weight should be taken through arms and legs (not torso leaning on bench)

0 = does not initiate cruising to right/left
1 = initiate cruising, < 1 complete step to right/left
2 = cruises 1 – 4 steps to right/left
3 = cruises 5 steps to right/left

Cruising one step = movement sideways of **both** legs

67/68. Standing two/one hand(s) held: Walks forward 10 steps
0 = does not initiate walking forward
1 = walks forward < 3 steps
2 = walks forward 1 – 9 steps
3 = walks forward 10 steps

One step is push off to contact of **one** leg
Steps must be consecutive, short break of 1-2 sec allowed

69. Standing: Walks forward 10 steps
Starting: must stand arms free. Can hold on beforehand but must balance in arms free before starting walk

0 = does not initiate walking forward
1 = walks forward < 3 steps
2 = walks forward 1 – 9 steps
3 = walks forward 10 steps

70 .Standing: Walks forward 10 steps, stops, turns 180°, returns
Starting: must stand arms free. Can hold on beforehand but must balance in arms free before starting walk

0 = walks forward 10 steps, does not stop without falling
1 = walks forward 10 steps, stops, does not initiate turn
2 = walks forward 10 steps, stops, turns < 180°
3 = walks forward 10 steps, stops, turns 180°, returns

Must make **full stop** before turning
71. Standing: Walks backwards 10 steps
Starting: must stand arms free. Can hold on beforehand but must balance in arms free before starting walk

0 = does not initiate walking backwards
1 = walks backwards < 3 steps
2 = walks backwards 1 – 9 steps
3 = walks backwards 10 steps

72. Standing: Walks forward 10 steps carrying large object with two hands
0 = does not initiate walking, carrying large object
1 = walks forward 10 steps carrying a small object with one hand
2 = walks forward 10 steps, carrying a small object with two hands
3 = walks forward 10 steps, carrying a large object with two hands

73. Standing: Walks forward 10 consecutive steps between parallel lines 20 cm apart
0 = does not initiate walking
1 = walks forward < 3 consec. steps between lines
2 = walks forward 3-9 consec. steps between lines
3 = walks forward 10 consec. steps between lines

Part of the foot may touch line but not go over it.

74. Standing: Walks forward 10 consecutive steps on a straight line 2 cm wide
0 = does not initiate walking
1 = walks forward < 3 consecutive steps on line
2 = walks forward 3-9 consecutive steps on line
3 = walks forward 10 consecutive steps on line

Part of the foot must stay on the line

75/76. Standing: Steps over a stick at knee level, right/left foot leading
0 = does not initiate stepping over stick
1 = steps over stick 5-7.5 cm high
2 = steps over stick at mid-calf level
3 = steps over stick at knee level, right/left foot leading

Must finish without falling

77. Standing: Runs 4.5 m, stops and returns
0 = does not initiate running
1 = initiates running by walking quickly
2 = runs <4.5 m
3 = runs 4.5 m, stops and returns

For running both feet must be off the floor at some point

75/76. Standing: Kicks ball with right/left foot
0 = does not initiate kicking
1 = lifts right foot, does not kick
2 = kicks ball with right foot, but falls
3 = kicks ball with right foot

Kick: foot must clear the floor when ball is contacted and ball must move from the impact

80. Standing: Jumps 30 cm high, both feet simultaneously
0 = does not initiate jump
1 = jumps < 5 cm high, both feet simultaneously
2 = jumps 5-28 cm high, both feet simultaneously
3 = jumps 30 cm high, both feet simultaneously

Both feet must be off the floor at the same time, but not necessarily land or take off at the same time
Must land, arms free, without falling to get any score

81. Standing: Jumps forward 30 cm, both feet simultaneously
0 = does not initiate jump
1 = jumps forward < 5 cm, both feet simultaneously
2 = jumps forward 5-28 cm, both feet simultaneously
3 = jumps forward 30 cm, both feet simultaneously

Both feet must be off the floor at the same time, but not necessarily land or take off at the same time
Must land, arms free, without falling to get any score

82/83. Standing: Hops on right/left foot 10 times within a 60 cm circle
0 = does not initiate hopping on right/left foot
1 = hops on right/left foot < 3 times within a 60 cm circle
2 = hops on right/left foot 3-9 times within a 60 cm circle
3 = hops on right/left foot 10 times within a 60 cm circle

Part of the foot must stay in the circle
Hopping foot must clear the floor and other foot must not touch floor
Not pause more than 2 sec between hops

84/85. Standing holding one rail: Walks up/down four steps, alternating feet, holding one rail
0 = does not initiate walking up steps, holding rail
1 = walks up/down 2 steps, holding rail, same foot leads
2 = walks up/down 4 steps, holding rail, alternating feet inconsistently
3 = walks up/down 4 steps, holding rail, alternating feet

86/87. Standing: Walks up/down four steps, alternating feet
0 = does not initiate walking up steps
1 = walks up/down 2 steps, same foot leads
2 = walks up/down 4 steps, alternating feet inconsistently
3 = walks up/down 4 steps, alternating feet

88. Standing on 15 cm step: Jumps off, both feet simultaneously
0 = does not initiate jumping off step
1 = jumps off, both feet simultaneously, but falls
2 = jumps off, both feet simultaneously, but uses hand to avoid falling
3 = jumps off, both feet simultaneously

Both feet simultaneously: both feet of ground at same time, but not necessarily leaving floor or landing at same time

Appendix 2FAQ

Functional Assessment Questionnaire

Mobility Levels
*Choose the **one** answer below that best describes your typical walking ability (**with the use of any needed assistive devices**).*
I...

1. Cannot take any steps at all.
2. Can do some stepping on my own with the help of another person. Do not take full weight on feet; do not walk on a routine basis.
3. Walk for exercise in therapy and less than typical household distances. Usually require assistance from another person.
4. Walk for household distances, but make slow progress. Do not use walking at home as preferred mobility (primarily walk in therapy).
5. Walk more than 15-50 feet but only indoors (walk for household distances).
6. Walk more than 15-50 feet outside the home, but usually use a wheelchair or buggy for community distances or in congested areas.
7. Walk outside the home for community distances, but only on level surfaces (cannot manage kerbs, uneven ground, or stairs without assistance of another person).
8. Walk outside the home for community distances, able to manage kerbs and uneven ground in addition to level surfaces, but usually require minimal assistance or supervision for safety.
9. Walk outside the home for community distances, easily get around on level surfaces, kerbs, and uneven ground, but have difficulty or require minimal assistance with running, climbing, and/or stairs.
10. Walk, run and climb on level and uneven ground without difficulty or assistance.

Higher Level Mobility Skills
Please tick all the things you are able to do:

- ☐ Walk carrying an object
- ☐ Walk carrying a fragile object or a glass of liquid
- ☐ Walk up and downstairs using the banister
- ☐ Walk up and downstairs without using the banister
- ☐ Step up and down a kerb independently
- ☐ Run
- ☐ Run well including around a corner with good control
- ☐ Take steps backwards
- ☐ Manoeuvre in tight areas
- ☐ Get on/off the bus independently
- ☐ Hop on the right foot
- ☐ Hop on the left foot
- ☐ Step over an object right foot first
- ☐ Step over an object left foot first
- ☐ Ride an escalator, stepping on/off by yourself

Appendix 3Canadian Occupational Performance Measure

2ND EDITION

authors: Mary Law, Sue Baptiste, Anne Carswell, Mary Ann McColl, Helene Polatajko, Nancy Pollock

The Canadian Occupational Performance Measure (COPM) is an individualized measure designed for use by occupational therapists to detect self-perceived change in occupational performance problems over time.

Step 1: IDENTIFICATION OF OCCUPATIONAL PERFORMANCE ISSUES

To identify occupational performance problems, concerns and issues, interview the client, asking about daily activities in self-care, productivity and leisure. Ask clients to identify daily activities which they want to do, need to do or are expected to do by encouraging them to think about a typical day. Then ask the client to identify which of these activities are difficult for them to do now to their satisfaction. Record these activity problems in Steps 1A, 1B or 1C.	Rating importance Using the scoring card provided ask the client to rate, on a scale of 1 to 10, the importance of each activity. Place the ratings in the corresponding boxes in Steps 1A, 1B, or 1C.
STEP 1A : Self Care _____ **Personal Care** _____ (dressing, bathing, hygiene) _____ **Functional Mobility** _____ (Indoor, outdoor, car) _____ **Community Management** _____ Public transport, shopping _____ Heavy bags _____ _____	**IMPORTANCE** ☐ ☐ ☐ ☐ ☐ ☐ ☐ ☐
Productivity **Paid/Unpaid Work** _____ (job, volunteering) _____ **Household Management** _____ (cleaning, cooking, laundry)_____	**IMPORTANCE** ☐ ☐ ☐ ☐

LEISURE	IMPORTANCE
Quiet Recreation (crafts, reading) _____ _____ _____	☐ ☐ ☐
Active Recreation (sports, outings, travel) _____ _____ _____	☐ ☐ ☐
Socialization (e.g. visits, parties) _____ _____ _____	☐ ☐ ☐

STEPS 3& 4 SCORING –INITIAL AND REASSESSMENT

Confirm with the client the 5 most important problems and record them below. Using the scoring cards, ask the client to rate each problem on performance and satisfaction, then calculate the total scores. Total scores are calculated by adding together the performance or satisfaction scores for all problems and dividing by the number of problems. At reassessment, the client scores each problem again for performance and satisfaction. Calculate the new scores and the change score.

Initial assessment: Occupational performance problems	performance 1	satisfaction 1	Reassessment: Performance2	Satisfaction2
1_____	☐	☐	☐	☐
2_____	☐	☐	☐	☐
3_____	☐	☐	☐	☐
4_____	☐	☐	☐	☐
5_____	☐	☐	☐	☐
SCORING: Total Score = Total performance or satisfaction scores / # of problems	Performance Score 1 ☐	Satisfaction Score 1 ☐	Performance Score 2 ☐	Satisfaction Score 2 ☐

Change in Performance= Performance Score 2 ☐ **-Performance Score 1** ☐ **=** ☐

Change in Satisfaction = Satisfaction Score 2 ☐ **-Satisfaction Score 1** ☐ **=** ☐

Your Health and Well-Being

This survey asks for your views about your health. This information will help keep track of how you feel and how well you are able to do your usual activities. ***Thank you for completing this survey!***

For each of the following questions, please tick the one box that best describes your answer.

1. In general, would you say your health is:

Excellent	Very good	Good	Fair	Poor
☐1	☐2	☐3	☐4	☐5

2. The following questions are about activities you might do during a typical day. Does <u>your health now limit you</u> in these activities? If so, how much?

	Yes, limited a lot	Yes, limited a little	No, not limited at all
a <u>Moderate activities</u>, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf	☐1	☐2	☐3
b Climbing <u>several</u> flights of stairs	☐1	☐2	☐3

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a Accomplished less than you would like	☐1	☐2	☐3	☐4	☐5
b Were limited in the kind of work or other activities	☐1	☐2	☐3	☐4	☐5

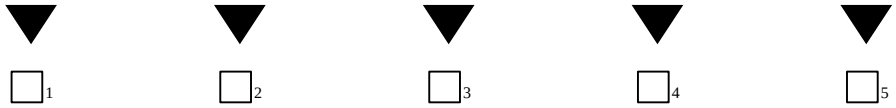
3. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of your physical health?

4. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a Accomplished less than you would like	☐1	☐2	☐3	☐4	☐5
b Did work or other activities less carefully than usual	☐1	☐2	☐3	☐4	☐5

5. During the past 4 weeks, how much did pain interfere with your normal work (including both work outside the home and housework)?

Not at all	A little bit	Moderately	Quite a bit	Extremely



6. These questions are about how you feel and how things have been with you <u>during the past 4 weeks</u>. For each question, please give the one answer that comes closest to the way you have been feeling. How much of the time during the <u>past 4 weeks</u>…

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a Have you felt calm and peaceful?	☐1	☐2	☐3	☐4	☐5
b Did you have a lot of energy?	☐1	☐2	☐3	☐4	☐5
c Have you felt downhearted and low?	☐1	☐2	☐3	☐4	☐5

7. During the <u>past 4 weeks</u>, how much of the time has your <u>physical health or emotional problems</u> interfered with your social activities (like visiting with friends, relatives, etc.)?

Appendix 5 Rosenberg Self-Esteem Scale

Subject ID:

Rosenberg Self-Esteem Scale

Instructions:
Below is a list of statements dealing with your general feelings about yourself. If you strongly agree, circle **SA**. If you agree with the statement, circle **A**. If you disagree, circle **D**. If you strongly disagree, circle **SD**.

1.	On the whole, I am satisfied with myself.	SA	A	D	SD
2.	At times, I think I am no good at all.	SA	A	D	SD
3.	I feel that I have a number of good qualities.	SA	A	D	SD
4.	I am able to do things as well as most other people.	SA	A	D	SD
5.	I feel I do not have much to be proud of.	SA	A	D	SD
6.	I certainly feel useless at times.	SA	A	D	SD
7.	I feel that I'm a person of worth, at least on an equal plane with others.	SA	A	D	SD
8.	I wish I could have more respect for myself.	SA	A	D	SD
9.	All in all, I am inclined to feel that I am a failure.	SA	A	D	SD
10.	I take a positive attitude toward myself.	SA	A	D	SD

Rosenberg Self-Esteem scoring

Rosenberg Self-Esteem Scale (Rosenberg, 1965)

The scale is a ten item Likert scale with items answered on a four point scale - from strongly agree to strongly disagree. The original sample for which the scale was developed consisted of 5,024 High School Juniors and Seniors from 10 randomly selected schools in New York State.

Instructions: Below is a list of statements dealing with your general feelings about yourself. If you strongly agree, circle **SA**. If you agree with the statement, circle **A**. If you disagree, circle **D**. If you strongly disagree, circle **SD**.

1.	On the whole, I am satisfied with myself.	SA	A	D	SD
2.*	At times, I think I am no good at all.	SA	A	D	SD
3.	I feel that I have a number of good qualities.	SA	A	D	SD
4.	I am able to do things as well as most other people.	SA	A	D	SD
5.*	I feel I do not have much to be proud of.	SA	A	D	SD
6.*	I certainly feel useless at times.	SA	A	D	SD
7.	I feel that I'm a person of worth, at least on an equal plane with others.	SA	A	D	SD
8.*	I wish I could have more respect for myself.	SA	A	D	SD
9.*	All in all, I am inclined to feel that I am a failure.	SA	A	D	SD
10.	I take a positive attitude toward myself.	SA	A	D	SD

Scoring: SA=3, A=2, D=1, SD=0. Items with an asterisk are reverse scored, that is, SA=0, A=1, D=2, SD=3. Sum the scores for the 10 items. The higher the score, the higher the self esteem.

The scale may be used without explicit permission. The author's family, however, would like to be kept informed of its use:

The Morris Rosenberg Foundation
c/o Department of Sociology
University of Maryland
2112 Art/Soc Building
College Park, MD 20742-1315

References

References with further characteristics of the scale:

Crandal, R. (1973). The measurement of self-esteem and related constructs, Pp. 80-82 in J.P. Robinson & P.R. Shaver (Eds), **Measures of social psychological attitudes. Revised edition**. Ann Arbor: ISR.

Appendix 6 Search strategies study 1 (systematic review, Chapter 4)

Database search*(last updated 14th January 2016)*

PubMed

#1 ('cerebral palsy')
"Cerebral Palsy"[mesh] OR "cerebral palsy" [tiab] OR "cerebral palsies" [tiab] OR "little disease"[tiab] OR "little's disease"[tiab] OR "spastic diplegias"[tiab] OR "spastic diplegia"[tiab]

#2 ('gait')
gait[tiab OR walk [tiab] OR "gait quality"[tiab] OR "walk test"[tiab] OR "walking performance"[tiab]

#3 ('outcome measure')
(test[ti] OR tests[ti] OR tool[ti] OR tools[ti] OR instrument[ti] OR instruments[ti] OR scale[ti]) AND (analys*[tiab] OR assess*[tiab] OR determin*[tiab] OR evaluat*[tiab] OR measure*[tiab] OR quantif*[tiab] OR score*[tiab]) or Analy*[ti] OR assessment*[tiab] OR assessing[tiab] OR evaluat*[ti] OR instrument[tiab] OR instruments[tiab] OR measure[tiab] OR measurement*[tiab] OR measures[tiab] OR quantifying[tiab] OR quantification[tiab] OR questionnaire*[tw] OR scale[tiab] OR scales[tiab] OR score[tiab] OR scores[tiab] OR screening[tiab] OR subtest*[tiab] OR test[tiab] OR tests[tiab] OR testing[tiab] OR tool[tiab] OR tools[tiab]

#4 ('sensitive filter for measurement properties') (Terwee et al 2009)
(instrumentation[sh] OR methods[sh] OR Validation Studies[pt] OR Comparative Study[pt] OR psychometrics [MeSH] OR psychometr*[tiab] OR clinimetr*[tw] OR clinometr*[tw] OR ''outcome assessment (health care)''[MeSH] OR outcome assessment[tiab] OR outcome measure*[tw] ''observer variation''[MeSH] OR observer variation[tiab] OR ''Health Status Indicators''[Mesh] OR ''reproducibility of results''[MeSH] OR reproducib*[tiab] OR ''discriminant analysis''[MeSH] OR reliab*[tiab] OR unreliab*[tiab] OR valid*[tiab] OR coefficient[tiab] OR homogeneity[tiab] OR homogeneous[tiab] OR ''internal consistency''[tiab] OR (cronbach*[tiab] AND (alpha[tiab] OR alphas[tiab])) OR (item[tiab] AND (correlation*[tiab] OR selection*[tiab] OR reduction*[tiab])) OR agreement[tiab] OR precision[tiab] OR imprecision[tiab] OR ''precise values''[tiab] OR test-retest[tiab] OR (test[tiab] AND retest[tiab]) OR (reliab*[tiab] AND (test[tiab] OR retest[tiab])) OR stability[tiab] OR interrater[tiab] OR inter-rater[tiab] OR intrarater[tiab] OR intra-rater[tiab] OR intertester[tiab] OR inter-tester[tiab] OR intratester[tiab] OR intra-tester[tiab] OR interobserver[tiab] OR inter-observer[tiab] OR intraobserver[tiab] OR intraobserver[tiab] OR intertechnician[tiab] OR inter-technician[tiab] OR intratechnician[tiab] OR intra-technician[tiab] OR interexaminer[tiab] OR inter-examiner[tiab] OR intraexaminer[tiab] OR intra-examiner[tiab] OR interassay[tiab] ORinter-assay[tiab] OR intraassay[tiab] OR intra-assay[tiab] OR interindividual[tiab] OR inter-individual[tiab] OR intraindividual[tiab] OR intra-individual[tiab] OR interparticipant[tiab] OR inter-participant[tiab] OR intraparticipant[tiab] OR intra-participant[tiab] OR kappa[tiab] OR kappa's[tiab] OR kappas[tiab] OR repeatab*[tiab] OR ((replicab*[tiab] OR repeated[tiab]) AND (measure[tiab] OR measures[tiab] OR findings[tiab] OR result[tiab] OR results[tiab] OR test[-tiab] OR tests[tiab])) OR generaliza*[tiab] OR generalisa*[tiab] OR concordance[tiab] OR (intraclass[tiab] AND correlation*[tiab]) OR discriminative[tiab] OR ''known

group''[tiab] OR factor analysis[tiab] OR factor analyses[tiab] OR dimension*[tiab] OR subscale*[tiab] OR (multitrait[tiab] AND scaling[tiab] AND (analysis[tiab] OR analyses[tiab])) OR item discriminant[tiab] OR interscale correlation*[tiab] OR error[tiab] OR errors[tiab] OR ''individual variability''[tiab] OR (variability[tiab] AND (analysis[tiab] OR values[tiab])) OR (uncertainty[tiab] AND (measurement[tiab] OR measuring[tiab])) OR ''standard error of measurement''[tiab] OR sensitiv*[tiab] OR responsive*[tiab] OR ((minimal[tiab] OR minimally[tiab] OR clinical[tiab] OR clinically[tiab]) AND (important[tiab] OR significant[tiab]
OR detectable[tiab])AND(change[tiab]ORdifference[tiab])) OR (small*[tiab] AND (real[tiab] OR detectable[tiab]) AND (change[tiab] OR difference[tiab])) OR meaningful change[tiab] OR ''ceiling effect''[tiab] OR ''floor effect''[tiab] OR ''Item response model''[tiab] OR IRT[tiab] OR Rasch[tiab] OR ''Differential item functioning''[tiab] OR DIF[tiab] OR ''computer adaptive testing''[tiab] OR ''item bank''[tiab] OR ''cross-cultural equivalence''[tiab])

#6 ('exclusion filter') (Terwee et al 2009)
(addresses OR biography OR case reports OR comment OR directory OR editorial OR festschrift OR interview OR lectures OR legal cases OR legislation OR letter OR news OR newspaper article OR patient education handout OR popular works OR congresses OR consensus development conference OR consensus development conference OR practice guideline) NOT (''animals''[MeSH Terms] NOT ''humans''[MeSH Terms])

Combination search
#1 AND #2 and #3 AND #4 = #5
#5 NOT #6

Medline

#1 ('cerebral palsy')
AB "Cerebral Palsy" OR AB "cerebral palsy" OR AB "cerebral palsies" OR "little's disease" OR AB "spastic diplegias" OR AB "spastic diplegia"

#2 ('gait')
AB gait OR AB walk OR AB "gait quality" OR AB "walk test" OR AB "walking performance"

#3 ('outcome measure')
AB test OR AB tests OR AB tool OR AB tools OR AB instrument OR AB instruments OR AB scale OR AB analys* OR AB assess* OR AB determin* OR AB evaluat* OR AB measure* OR AB quantif*[tiab] OR AB score*or AB Analy* OR AB assessment* OR AB assessing OR AB evaluat* OR AB instrument OR AB instruments OR AB measure OR AB measurement* OR AB measures OR AB quantifying OR AB quantification OR AB questionnaire* OR AB scale OR AB scales OR AB score OR AB scores OR AB screening OR AB subtest* OR AB test OR AB tests OR AB testing OR AB tool OR AB tools

#4 ('properties')
AB accuracy OR AB accurate OR AB clinimetr* OR AB coefficient* OR AB consisten* OR AB correlated OR AB correlation* OR AB cronbach OR AB discrimina* OR AB interrater OR AB inter-rater OR AB intersession OR AB inter-session OR AB intertester OR AB inter-tester OR AB Intrarater OR AB intra-rater OR AB intratester OR AB intra-tester OR AB kappa OR AB Observer variationOR AB predictiv* OR AB propert* OR AB Psychometrics OR AB psychometr* OR AB reliab* OR AB repeatable OR AB repeatability

OR AB "Reproducibility of Results" OR AB reproducible OR AB reproducibility OR AB responsive* OR AB "Sensitivity and Specificity" OR AB sensitive OR AB sensitivity OR AB spearman* OR AB specific OR AB specificity OR AB spearman OR AB subscale* OR AB suitable OR AB suitability OR AB "test development" OR AB test-retest OR AB useful* OR AB utility OR AB valid OR AB validity OR AB validat* OR AB "Validation studies"

#6 ('exclusion filter')
AB biography OR AB case reports OR AB comment OR AB directory OR AB editorial OR AB interview OR AB lectures OR AB legal cases OR AB legislation OR AB letter OR AB news OR AB newspaper article OR AB patient education handout OR popular works OR congresses OR consensus development conference OR consensus development conference OR practice guideline) OR AB Stroke OR AB Parkinson disease OR AB stroke OR AB parkinson* NOT (''animals'')

Combination search
#1 AND #2 AND #3 AND #4 = #5
#5 NOT #6

CINAHL

#1 ('cerebral palsy')
AB "Cerebral Palsy" OR AB "cerebral palsy" OR AB "cerebral palsies" OR "little's disease" OR AB "spastic diplegias" OR AB "spastic diplegia"

#2 ('gait')
AB gait OR AB walk OR AB "gait quality" OR AB "walk test" OR AB "walking performance"

#3 ('outcome measure')
AB test OR AB tests OR AB tool OR AB tools OR AB instrument OR AB instruments OR AB scale OR AB analys* OR AB assess* OR AB determin* OR AB evaluat* OR AB measure* OR AB quantif*[tiab] OR AB score*or AB Analy* OR AB assessment* OR AB assessing OR AB evaluat* OR AB instrument OR AB instruments OR AB measure OR AB measurement* OR AB measures OR AB quantifying OR AB quantification OR AB questionnaire* OR AB scale OR AB scales OR AB score OR AB scores OR AB screening OR AB subtest* OR AB test OR AB tests OR AB testing OR AB tool OR AB tools

#4 ('properties')
AB accuracy OR AB accurate OR AB clinimetr* OR AB coefficient* OR AB consisten* OR AB correlated OR AB correlation* OR AB cronbach OR AB discrimina* OR AB interrater OR AB inter-rater OR AB intersession OR AB inter-session OR AB intertester OR AB inter-tester OR AB Intrarater OR AB intra-rater OR AB intratester OR AB intra-tester OR AB kappa OR AB Observer variationOR AB predictiv* OR AB propert* OR AB Psychometrics OR AB psychometr* OR AB reliab* OR AB repeatable OR AB repeatability OR AB "Reproducibility of Results" OR AB reproducible OR AB reproducibility OR AB responsive* OR AB "Sensitivity and Specificity" OR AB sensitive OR AB sensitivity OR AB spearman* OR AB specific OR AB specificity OR AB spearman OR AB subscale* OR AB suitable OR AB suitability OR AB "test development" OR AB test-retest OR AB useful* OR AB utility OR AB valid OR AB validity OR AB validat* OR AB "Validation studies"

#6 ('exclusion filter')

AB biography OR AB case reports OR AB comment OR AB directory OR AB editorial OR AB interview OR AB lectures OR AB legal cases OR AB legislation OR AB letter OR AB news OR AB newspaper article OR AB patient education handout OR popular works OR congresses OR consensus development conference OR consensus development conference OR practice guideline) OR AB Stroke OR AB Parkinson disease OR AB stroke OR AB parkinson* NOT (''animals'')

Combination search
#1 AND #2 AND #3 AND #4 = #5
#5 NOT #6

Scopus

#1 ('cerebral palsy')
(TITLE-ABS-KEY (cerebral palsy) OR (cerebral palsies) OR (spastic diplegias)OR (spastic diplegia))

#2 ('gait')
(TITLE-ABS-KEY (gait) OR TITLE-ABS-KEY (walk) TITLE-ABS-KEY (gait quality*) OR TITLE-ABS-KEY (walk test*) OR TITLE-ABS-KEY (walking performance))

#3 ('outcome measure')
(TITLE-ABS-KEY(test) OR TITLE-ABS-KEY (tests) OR TITLE-ABS-KEY (tool) OR TITLE-ABS-KEY (tools) OR TITLE-ABS-KEY (instrument) OR TITLE-ABS-KEY (instruments) OR TITLE-ABS-KEY (scale) OR TITLE-ABS-KEY (analys*) OR TITLE-ABS-KEY (assess*) OR TITLE-ABS-KEY (determin*) OR TITLE-ABS-KEY (evaluat*) OR TITLE-ABS-KEY (measure*) OR TITLE-ABS-KEY (quantif*) OR TITLE-ABS-KEY score*) OR TITLE-ABS-KEY (Analy*) OR TITLE-ABS-KEY (assessment*) OR TITLE-ABS-KEY (assessing) OR TITLE-ABS-KEY (evaluat*) OR TITLE-ABS-KEY (instrument) OR TITLE-ABS-KEY (instruments) OR TITLE-ABS-KEY (measure) OR TITLE-ABS-KEY (measurement*) OR TITLE-ABS-KEY (measures) OR TITLE-ABS-KEY (quantifying) OR TITLE-ABS-KEY (quantification) OR TITLE-ABS-KEY (questionnaire*) OR TITLE-ABS-KEY (scale) OR TITLE-ABS-KEY (scales) OR TITLE-ABS-KEY (score) OR TITLE-ABS-KEY (scores) OR TITLE-ABS-KEY (screening) OR TITLE-ABS-KEY (subtest*) OR TITLE-ABS-KEY (test) OR TITLE-ABS-KEY (tests) OR TITLE-ABS-KEY (testing) OR TITLE-ABS-KEY (tool) OR TITLE-ABS-KEY (tools))

Combination search
#1 AND #2 AND #3

Appendix 7 COSMIN checklist

COSMIN checklist with 4-point scale

Contact
CB Terwee, PhD
VU University Medical Center
Department of Epidemiology and Biostatistics
EMGO Institute for Health and Care Research
1081 BT Amsterdam
The Netherlands
Website: www.cosmin.nl, www.emgo.nl
E-mail: cb.terwee@vumc.nl

Instructions

This version of the COSMIN checklist is recommended for use in systematic reviews of measurement properties. With this version it is possible to calculate overall methodological quality scores per study on a measurement property. A methodological quality score per box is obtained by taking the lowest rating of any item in a box ('worse score counts'). For example, if for a reliability study one item in the box 'Reliability' is scored poor, the methodological quality of that reliability study is rated as poor. The Interpretability box and the Generalizability box are mainly used as data extraction forms. We recommend to use the Interpretability box to extract all information on the interpretability issues described in this box (e.g. norm scores, floor-ceiling effects, minimal important change) of the instruments under study from the included articles. Similar, we recommend to use the Generalizability box to extract data on the characteristics of the study population and sampling procedure. Therefore no scoring system was developed for these boxes.

This scoring system is described in this paper:

Terwee CB, Mokkink LB, Knol DL, Ostelo RWJG, Bouter LM, de Vet HCW. Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist. Quality of Life Research 2011, July 6 [epub ahead of print].

Box B. Reliability: relative measures (including test-retest reliability, inter-rater reliability and intra-rater reliability)				
	excellent	**good**	**fair**	**poor**
Design requirements				
1 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3 Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4 Were at least two measurements available?	At least two measurements			Only one measurement
5 Were the administrations independent?	Independent measurements	Assumable that the measurements were independent	Doubtful whether the measurements were independent	measurements NOT independent
6 Was the time interval stated?	Time interval stated		Time interval NOT stated	
7 Were patients stable in the interim period on the construct to be measured?	Patients were stable (evidence provided)	Assumable that patients were stable	Unclear if patients were stable	Patients were NOT stable
8 Was the time interval appropriate?	Time interval appropriate		Doubtful whether time interval was appropriate	Time interval NOT appropriate

9	Were the test conditions similar for both measurements? e.g. type of administration, environment, instructions	Test conditions were similar (evidence provided)	Assumable that test conditions were similar	Unclear if test conditions were similar	Test conditions were NOT similar
10	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
11	for continuous scores: Was an intraclass correlation coefficient (ICC) calculated?	ICC calculated and model or formula of the ICC is described	ICC calculated but model or formula of the ICC not described or not optimal. Pearson or Spearman correlation coefficient calculated with evidence provided that no systematic change has occurred	Pearson or Spearman correlation coefficient calculated WITHOUT evidence provided that no systematic change has occurred or WITH evidence that systematic change has occurred	No ICC or Pearson or Spearman correlations calculated
12	for dichotomous/nominal/ordinal scores: Was kappa calculated?	Kappa calculated			Only percentage agreement calculated
13	for ordinal scores: Was a weighted kappa calculated?	Weighted Kappa calculated		Unweighted Kappa calculated	Only percentage agreement calculated
14	for ordinal scores: Was the weighting scheme described? e.g. linear, quadratic	Weighting scheme described	Weighting scheme NOT described		

Box C. Measurement error: absolute measures				
	excellent	**good**	**fair**	**poor**
Design requirements				
1 Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2 Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3 Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4 Were at least two measurements available?	At least two measurements			Only one measurement
5 Were the administrations independent?	Independent measurements	Assumable that the measurements were independent	Doubtful whether the measurements were independent	measurements NOT independent
6 Was the time interval stated?	Time interval stated		Time interval NOT stated	
7 Were patients stable in the interim period on the construct to be measured?	Patients were stable (evidence provided)	Assumable that patients were stable	Unclear if patients were stable	Patients were NOT stable
8 Was the time interval appropriate?	Time interval appropriate		Doubtful whether time interval was appropriate	Time interval NOT appropriate

9	Were the test conditions similar for both measurements? e.g. type of administration, environment, instructions	Test conditions were similar (evidence provided)	Assumable that test conditions were similar	Unclear if test conditions were similar	Test conditions were NOT similar
10	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
11	for CTT: Was the Standard Error of Measurement (SEM), Smallest Detectable Change (SDC) or Limits of Agreement (LoA) calculated?	SEM, SDC, or LoA calculated	Possible to calculate LoA from the data presented		SEM calculated based on Cronbach's alpha, or on SD from another population

Box D. Content validity (including face validity)					
		excellent	**good**	**fair**	**poor**
General requirements					
1	Was there an assessment of whether all items refer to relevant aspects of the construct to be measured?	Assessed if all items refer to relevant aspects of the construct to be measured		Aspects of the construct to be measured poorly described AND this was not taken into consideration	NOT assessed if all items refer to relevant aspects of the construct to be measured
2	Was there an assessment of whether all items are relevant for the study population? (e.g. age, gender, disease characteristics, country, setting)	Assessed if all items are relevant for the study population in adequate sample size (≥10)	Assessed if all items are relevant for the study population in moderate sample size (5-9)	Assessed if all items are relevant for the study population in small sample size (<5)	NOT assessed if all items are relevant for the study population OR target population not involved
3	Was there an assessment of whether all items are relevant for the purpose of the measurement instrument? (discriminative, evaluative, and/or predictive)	Assessed if all items are relevant for the purpose of the application	Purpose of the instrument was not described but assumed	NOT assessed if all items are relevant for the purpose of the application	
4	Was there an assessment of whether all items together comprehensively reflect the construct to be measured?	Assessed if all items together comprehensively reflect the construct to be measured		No theoretical foundation of the construct and this was not taken into consideration	NOT assessed if all items together comprehen-sively reflect the construct to be measured
5	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study

Box F. Hypotheses testing					
		excellent	**good**	**fair**	**Poor**
Design requirements					
1	Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2	Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3	Was the sample size included in the analysis adequate?	Adequate sample size (≥100 per analysis)	Good sample size (50-99 per analysis)	Moderate sample size (30-49 per analysis)	Small sample size (<30 per analysis)

4	Were hypotheses regarding correlations or mean differences formulated a priori (i.e. before data collection)?	Multiple hypotheses formulated a priori	Minimal number of hypotheses formulate a priori	Hypotheses vague or not formulated but possible to deduce what was expected	Unclear what was expected
5	Was the expected *direction* of correlations or mean differences included in the hypotheses?	Expected direction of the correlations or differences stated	Expected direction of the correlations or differences NOT stated		
6	Was the expected absolute or relative *magnitude* of correlations or mean differences included in the hypotheses?	Expected magnitude of the correlations or differences stated	Expected magnitude of the correlations or differences NOT stated		
7	for convergent validity: Was an adequate description provided of the comparator instrument(s)?	Adequate description of the constructs measured by the comparator instrument(s)	Adequate description of most of the constructs measured by the comparator instrument(s)	Poor description of the constructs measured by the comparator instrument(s)	NO description of the constructs measured by the comparator instrument(s)
8	for convergent validity: Were the measurement properties of the comparator instrument(s) adequately described?	Adequate measurement properties of the comparator instrument(s) in a population similar to the study population	Adequate measurement properties of the comparator instrument(s) but not sure if these apply to the study population	Some information on measurement properties (or a reference to a study on measurement properties) of the comparator instrument(s) in any study population	No information on the measurement properties of the comparator instrument(s)

9	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study (e.g. only data presented on a comparison with an instrument that measures another construct)	Other important methodological flaws in the design or execution of the study
Statistical methods					
10	Were design and statistical methods adequate for the hypotheses to be tested?	Statistical methods applied appropriate	Assumable that statistical methods were appropriate, e.g. Pearson correlations applied, but distribution of scores or mean (SD) not presented	Statistical methods applied NOT optimal	Statistical methods applied NOT appropriate

Box H. Criterion validity					
		excellent	**good**	**fair**	**poor**
Design requirements					
1	Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2	Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3	Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4	Can the criterion used or employed be considered as a reasonable 'gold standard'?	Criterion used can be considered an adequate 'gold standard' (evidence provided)	No evidence provided, but assumable that the criterion used can be considered an adequate 'gold standard'	Unclear whether the criterion used can be considered an adequate 'gold standard'	Criterion used can NOT be considered an adequate 'gold standard'

5	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study	Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods				
6	for continuous scores: Were correlations, or the area under the receiver operating curve calculated?	Correlations or AUC calculated		Correlations or AUC NOT calculated
7	for dichotomous scores: Were sensitivity and specificity determined?	Sensitivity and specificity calculated		Sensitivity and specificity NOT calculated

Box I. Responsiveness					
		excellent	**good**	**fair**	**poor**
Design requirements					
1	Was the percentage of missing items given?	Percentage of missing items described	Percentage of missing items NOT described		
2	Was there a description of how missing items were handled?	Described how missing items were handled	Not described but it can be deduced how missing items were handled	Not clear how missing items were handled	
3	Was the sample size included in the analysis adequate?	Adequate sample size (≥100)	Good sample size (50-99)	Moderate sample size (30-49)	Small sample size (<30)
4	Was a longitudinal design with at least two measurement used?	Longitudinal design used			No longitudinal design used
5	Was the time interval stated?	Time interval adequately described			Time interval NOT described
6	If anything occurred in the interim period (e.g. intervention, other relevant events), was it adequately described?	Anything that occurred during the interim period (e.g. treatment) adequately described	Assumable what occurred during the interim period	Unclear or NOT described what occurred during the interim period	
7	Was a proportion of the patients changed (i.e. improvement or deterioration)?	Part of the patients were changed (evidence provided)	NO evidence provided, but assumable that part of the patients were changed	Unclear if part of the patients were changed	Patients were NOT changed
Design requirements for hypotheses testing					
	For constructs for which a gold standard was not available:				
8	Were hypotheses about changes in scores formulated a priori (i.e. before data collection)?	Hypotheses formulated a priori		Hypotheses vague or not formulated but possible to deduce what was expected	Unclear what was expected
9	Was the expected *direction* of correlations or mean differences of the change scores of HR-PRO instruments included in these hypotheses?	Expected direction of the correlations or differences stated	Expected direction of the correlations or differences NOT stated		
10	Were the expected absolute or relative *magnitude* of correlations or mean differences of the change scores of HR-PRO instruments included in these hypotheses?	Expected magnitude of the correlations or differences stated	Expected magnitude of the correlations or differences NOT stated		

11	Was an adequate description provided of the comparator instrument(s)?	Adequate description of the constructs measured by the comparator instrument(s)		Poor description of the constructs measured by the comparator instrument(s)	NO description of the constructs measured by the comparator instrument(s)
12	Were the measurement properties of the comparator instrument(s) adequately described?	Adequate measurement properties of the comparator instrument(s) in a population similar to the study population	Adequate measurement properties of the comparator instrument(s) but not sure if these apply to the study population	Some information on measurement properties (or a reference to a study on measurement properties) of the comparator instrument(s) in any study population	NO information on the measurement properties of the comparator instrument(s)
13	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study (e.g. only data presented on a comparison with an instrument that measures another construct)	Other important methodological flaws in the design or execution of the study
Statistical methods					
14	Were design and statistical methods adequate for the hypotheses to be tested?	Statistical methods applied appropriate		Statistical methods applied NOT optimal	Statistical methods applied NOT appropriate

Design requirement for comparison to a gold standard					
	For constructs for which a gold standard was available:				
15	Can the criterion for change be considered as a reasonable gold standard?	Criterion used can be considered an adequate 'gold standard' (evidence provided)	No evidence provided, but assumable that the criterion used can be considered an adequate 'gold standard'	Unclear whether the criterion used can be considered an adequate 'gold standard'	Criterion used can NOT be considered an adequate 'gold standard'
16	Were there any important flaws in the design or methods of the study?	No other important methodological flaws in the design or execution of the study		Other minor methodological flaws in the design or execution of the study	Other important methodological flaws in the design or execution of the study
Statistical methods					
17	for continuous scores: Were correlations between change scores, or the area under the Receiver Operator Curve (ROC) curve calculated?	Correlations or Area under the ROC Curve (AUC) calculated			Correlations or AUC NOT calculated
18	for dichotomous scales: Were sensitivity and specificity (changed versus not changed) determined?	Sensitivity and specificity calculated			Sensitivity and specificity NOT calculated

Appendix 8 NHS & QMU ethics

University Hospitals Division

NHS Lothian

Queen's Medical Research Institute
47 Little France Crescent, Edinburgh, EH16 4TJ

RESEARCH & DEVELOPMENT
Room E1.12
Tel: 0131 242 3330
Email:
R&DOffice@nhslothian.scot.nhs.uk

Director:
Professor David E Newby

SS/GR/LG

8th September 2014

Dr Marietta L Van Der Linden
Senior Research Fellow Physiotherapy
Queen Margaret University Drive
Musselburgh
Edinburgh
EH21 6UU

Dear Dr Van Der Linden

REC No:	11/AL/0044
R&D Project ID No:	2011/C/PT/01
Amendment:	Minor amendment dated 6th August 2014
Title of Research	The effects of a pragmatic community-based exercise therapy intervention on physical fitness, habitual physical activity, self-esteem and quality of life in young people with cerebral palsy, a pilot study.

I am writing in reply to recent correspondence in relation to an amendment(s) to the above project.

- Addition of Researcher – Asfarina Zanudin

We have now assessed any consequential changes and can confirm that NHS Lothian management approval is extended to cover the specific changes intimated.

Yours sincerely

p.p. G Rabelon

Mrs Susan Shepherd
Head of Research Governance

cc: Pamela Shand, NRS

Lothian NHS Board

Waverley Gate
2-4 Waterloo Place
Edinburgh
EH1 3EG
Telephone 0131 536 9000
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www.nhslothian.scot.nhs.uk

Dr Marietta van der Linden
Research Fellow
Queen Margaret university drive
Musselburgh
EH12 6UU

07 April 2011

Dear Dr van der Linden,

Study title: **The effects of a pragmatic community-based exercise therapy intervention on physical fitness, habitual physical activity, self-esteem and quality of life in young people with cerebral palsy, a pilot study.**
REC reference number: **11/AL/0044**
SSA reference number: **11/AL/0207**

The REC gave a favourable ethical opinion to this study.

Notification(s) have been received from local assessor(s), following site-specific assessment. On behalf of the Committee, I am pleased to confirm the extension of the favourable opinion to the new site(s) and investigator(s) listed below:

Research Site	Principal Investigator
Queen Margaret University/Edinburgh Leisure	Dr Marietta van der Linden

The favourable opinion is subject to management permission or approval being obtained from the host organisation prior to the start of the study at the site concerned.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

11/AL/0044	***Please quote this number on all correspondence***

Yours sincerely,

Dr Alex Bailey
Committee Co-ordinator
Email: alex.bailey@lhb.scot.nhs.uk

DIVISION OF DIETETICS, NUTRITION, BIOLOGICAL SCIENCES, PHYSIOTHERAPY, PODIATRY and RADIOGRAPHY

PROJECT RELEASE FORM resubmission

This form is designed to notify students and staff of the DivREC response to individual research project proposals and Dissertation Proposals. A copy of the form will also be retained by the Committee to record each decision and to monitor resource requirements.*Students must complete sections a – e below, otherwise you will be asked to re-submit:*

a. PROJECT TITLE: Identification of the reliability of physical function outcome measures in young people with CP and age-matched healthy controls, through a test-retest study design

b. STUDENT(S): Asfarina Zanudin 09004220

Please add in all names and matriculation numbers for group projects

c. SUPERVISOR: Dr Marietta van der Linden

d. SITE FOR DATA COLLECTION: Motion Gait Lab & Sport Hall, QMU

(If not QMU state where)

e. APPROXIMATE DATES FOR DATA COLLECTION: 30/9/2014-31/7/2015

All students should refer to Committee Response below and Comments overleaf

COMMITTEE RESPONSE

Decision	**✓ / X**	**Date**
1. Project proposal and Ethical approval granted	✓	**1/10/14**
2. Proceed with minor modifications to the project proposal (*as noted in response overleaf*)		
3. Resubmit revised proposal by		
4. Resubmit revised ethics by *(insert date ……)*		
5. Submit for further ethics scrutiny (QMU / external)		
6. Project documentation incomplete		
7. Other …		

Please note – you cannot proceed to dissertation unless response box 1 and/or box 2 ticked (✓)

GENERAL AND SPECIFIC COMMENTS ARE PROVIDED OVERLEAF:

Issues raised from original submission now addressed

Note – disclosure received separate to Ethics application (email 19/08/2014)

Please note – minor changes to stated methodology should be discussed with project supervisor.

Any major changes to stated methodology must be notified to subject area DivREC representative and re-submitted to DivREC hubsite on the appropriate amendment form

This form will only be signed by Head of Division once project and ethical approval granted

Signature DiVREC member: ____Gillian Baer __derek Santos_____
DATE__01/10/2014_____

Appendix 9 Consent form

Queen Margaret University

EDINBURGH

Centre Number:
Study Number:

CONSENT FORM

Study Title:**The effects of exercise therapy on young people with cerebral palsy**

Name of Researcher: Asfarina Zanudin

Please initial box

1. I confirm that I have read and understand the information sheet dated Jan 2015 (version 3) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being ☐

3. I understand that sections of my medical notes and data collected during the study may be looked at by responsible individuals from Queen Margaret University, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records. ☐

4. I agree to my GP being informed on my participation in the study (optional) ☐

5. I agree to my picture and video being recorded for the purpose of this study ☐

6. I agree to take part in this study ☐

_____	_____	_____
Name of Patient	Date	Signature
_____	_____	_____
Name of Person taking consent	Date	Signature

When completed 1 for patient; 1 for researcher site file; 1 (original) to be kept in medical notes

Appendix 10 Data collection sheet

Patient Identification Number: Date:

ActivPal Number:

Date of Birth:

Sex: Male / Female

GMFCS:

Walking aids:

Previous operations:

Co-morbidities:

Notes:

Did Canadian Occupational Performance Measure? Yes / No

TIMED UP AND GO (TUG) TEST

Walking aid used? Yes No

If yes, what aid was used? _____

Attempt1 _____seconds

Attempt2 _____seconds

Attempt2 _____seconds

Average_____seconds

RANGE OF MOTION

	Left	**Right**
Hip flexion, knee flexed		
Popliteal angle, opp knee flexed		
Knee extension		
Knee flexion		
Dorsiflexion, knee flexed		
Plantarflexion, knee flexed		

Hip Extension		

ANTHROPOMETRIC DATA:

Height (mm):

Weight:

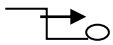
Limb Length (mm): Right _____ Left_____

Knee Width (mm): Right _____ Left_____

Malleolar Width (mm): Right _____ Left_____

Tibial Torsion: Right _____ Left_____

MUSCLE STRENGTH:

Muscle Group	Side	Lever arm length (mm)	Force		
			Trial 1	Trial 2	Trial 3
Hip Extensors 	Right				
	Left				
Hip Abductors	Right				
	Left				
Knee Extensors 	Right				
	Left				

Vicon directory _____

_____

Comments/Walking aids etc_____

_____

_____

_____

GAIT ANALYSIS

Trial	#	Left Force plate	Right Force plate	Processed	Additional Info
Static					
Static					
Static					
Balance Right					
Balance Right					
Balance Right					
Balance Right					
Balance Left					
Balance Left					
Balance Left					
Balance Left					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					

Walk					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					
Walk					

GROSS MOTOR FUNCTION MEASURE

Evaluator's Name _____ GMFCS Level:_____

_____

Testing Conditions (e.g.: room, clothing, time, others present):_____

_____

_____

_____

_____

_____

_____

The GMFM is a standardized observational instrument designed and validated to measure change in gross motor function over time in children with cerebral palsy. The scoring key is meant to be a general guideline. However, most of the items have specific descriptors for each score. It is imperative that the guidelines contained in the manual be used for scoring each item.

SCORING KEY
0 = does not initiate
1 = initiates
2 = partially completes
3 = completes
NT = Not tested [used for the GMAE scoring*]

It is now important to differentiate a true score of "0" (child does not initiate) from an item which is Not Tested (NT) if you are interested in using the GMFM-66 Ability Estimator Software.

*The GMFM-66 Gross Motor Ability Estimator (GMAE) software is available with the GMFM manual (2002). The advantage of the software is the conversion of the ordinal scale into an interval scale. This will allow for a more accurate estimate of the child's ability and provide a measure that is equally responsive to change across the spectrum of ability levels. Items that are used in the calculation of the GMFM-66 score are shaded and identified with an asterisk (*). The GMFM-66 is only valid for use with children who have cerebral palsy.

Item		D: STANDING	SCORE				NT
*	52.	ON THE FLOOR: PULLS TO STD AT LARGE BENCH	0 ☐	1 ☐	2 ☐	3 ☐	52.
*	53.	STD: MAINTAINS, ARMS FREE, 3 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	53.
*	54.	STD: HOLDING ON TO LARGE BENCH WITH ONE HAND, LIFTS R FOOT, 3 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	54.
*	55.	STD: HOLDING ON TO LARGE BENCH WITH ONE HAND, LIFTS L FOOT, 3 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	55.
*	56.	STD: MAINTAINS, ARMS FREE, 20 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	56.
*	57.	STD: LIFTS L FOOT, ARMS FREE, 10 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	57.
*	58.	STD: LIFTS R FOOT, ARMS FREE, 10 SECONDS	0 ☐	1 ☐	2 ☐	3 ☐	58.
*	59.	SIT ON SMALL BENCH: ATTAINS STD WITHOUT USING ARMS	0 ☐	1 ☐	2 ☐	3 ☐	59.
*	60.	HIGH KN: ATTAINS STD THROUGH HALF KN ON R KNEE, WITHOUT USING ARMS	0 ☐	1 ☐	2 ☐	3 ☐	60.
*	61.	HIGH KN: ATTAINS STD THROUGH HALF KN ON L KNEE, WITHOUT USING ARMS	0 ☐	1 ☐	2 ☐	3 ☐	61.
*	62.	STD: LOWERS TO SIT ON FLOOR WITH CONTROL, ARMS FREE	0 ☐	1 ☐	2 ☐	3 ☐	62.
*	63.	STD: ATTAINS SQUAT, ARMS FREE	0 ☐	1 ☐	2 ☐	3 ☐	63.
*	64.	STD: PICKS UP OBJECT FROM FLOOR, ARMS FREE, RETURNS TO STAND	0 ☐	1 ☐	2 ☐	3 ☐	64.

TOTAL DIMENSION D ☐

Item		E: WALKING, RUNNING & JUMPING	SCORE				NT
*	65.	STD, 2 HANDS ON LARGE BENCH: CRUISES 5 STEPS TO R	0 ☐	1 ☐	2 ☐	3 ☐	65.
*	66.	STD, 2 HANDS ON LARGE BENCH: CRUISES 5 STEPS TO L	0 ☐	1 ☐	2 ☐	3 ☐	66.
*	67.	STD, 2 HANDS HELD: WALKS FORWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	67.
*	68.	STD, 1 HAND HELD: WALKS FORWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	68.
*	69.	STD: WALKS FORWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	69.
*	70.	STD: WALKS FORWARD 10 STEPS, STOPS, TURNS 180°, RETURNS	0 ☐	1 ☐	2 ☐	3 ☐	70.
*	71.	STD: WALKS BACKWARD 10 STEPS	0 ☐	1 ☐	2 ☐	3 ☐	71.
*	72.	STD: WALKS FORWARD 10 STEPS, CARRYING A LARGE OBJECT WITH 2 HANDS	0 ☐	1 ☐	2 ☐	3 ☐	72.
*	73.	STD: WALKS FORWARD 10 CONSECUTIVE STEPS BETWEEN PARALLEL LINES 20cm (8") APART	0 ☐	1 ☐	2 ☐	3 ☐	73.
*	74.	STD: WALKS FORWARD 10 CONSECUTIVE STEPS ON A STRAIGHT LINE 2cm (3/4") WIDE	0 ☐	1 ☐	2 ☐	3 ☐	74.
*	75.	STD: STEPS OVER STICK AT KNEE LEVEL, R FOOT LEADING	0 ☐	1 ☐	2 ☐	3 ☐	75.
*	76.	STD: STEPS OVER STICK AT KNEE LEVEL, L FOOT LEADING	0 ☐	1 ☐	2 ☐	3 ☐	76.
*	77.	STD: RUNS 4.5m (15'), STOPS & RETURNS	0 ☐	1 ☐	2 ☐	3 ☐	77.
*	78.	STD: KICKS BALL WITH R FOOT	0 ☐	1 ☐	2 ☐	3 ☐	78.
*	79.	STD: KICKS BALL WITH L FOOT	0 ☐	1 ☐	2 ☐	3 ☐	79.
*	80.	STD: JUMPS 30cm (12") HIGH, BOTH FEET SIMULTANEOUSLY	0 ☐	1 ☐	2 ☐	3 ☐	80.
*	81.	STD: JUMPS FORWARD 30 cm (12"), BOTH FEET SIMULTANEOUSLY	0 ☐	1 ☐	2 ☐	3 ☐	81.
*	82.	STD ON R FOOT: HOPS ON R FOOT 10 TIMES WITHIN A 60cm (24") CIRCLE	0 ☐	1 ☐	2 ☐	3 ☐	82.
*	83.	STD ON L FOOT: HOPS ON L FOOT 10 TIMES WITHIN A 60cm (24") CIRCLE	0 ☐	1 ☐	2 ☐	3 ☐	83.
*	84.	STD, HOLDING 1 RAIL: WALKS UP 4 STEPS, HOLDING 1 RAIL, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	84.
*	85.	STD, HOLDING 1 RAIL: WALKS DOWN 4 STEPS, HOLDING 1 RAIL, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	85.
*	86.	STD: WALKS UP 4 STEPS, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	86.
*	87.	STD: WALKS DOWN 4 STEPS, ALTERNATING FEET	0 ☐	1 ☐	2 ☐	3 ☐	87.
*	88.	STD ON 15cm (6") STEP: JUMPS OFF, BOTH FEET SIMULTANEOUSLY	0 ☐	1 ☐	2 ☐	3 ☐	88.

TOTAL DIMENSION E ☐

Was this assessment indicative of this child's 'regular' performance? Yes / No

COMMENTS:

AEROBIC FITNESS

GMFCS: I II III

Shuttle run test Level: _____ Shuttle: _____

Level 0 12 3 4 5 6 78

Level 1 12 3 4 5 6 78 9

Level 2 12 3 4 5 6 78 9

Level 3 12 3 4 5 6 78 9 10

Level 4 12 3 4 5 6 78 9 10

Level 5 12 3 4 5 6 78 9 10

Level 6 12 3 4 5 6 78 9 10 11

Level 7 12 3 4 5 6 78 9 10 11

Level 8 12 3 4 5 6 78 9 10 11 12

Level 9 12 3 4 5 6 78 9 10 11 12

Level 10 12 3 4 5 6 78 9 10 11 12

Level 11 12 3 4 5 6 78 9 10 11 12 13

Level 12 12 3 4 5 6 78 9 10 11 12 13

Level 13 12 3 4 5 6 78 9 10 11 12 13 14

Level 14 12 3 4 5 6 78 9 10 11 12 13 14

Level 15 12 3 4 5 6 78 9 10 11 12 13 14 15

Level 16 12 3 4 5 6 78 9 10 11 12 13 14 15

Level 17 12 3 4 5 6 78 9 10 11 12 13 14 15

Level 18 12 3 4 5 6 78 9 10 11 12 13 14 15

Level 19 12 3 4 5 6 78 9 10 11 12 13 14 15 16

Level 20 12 3 4 5 6 78 9 10 11 12 13 14 15 16 17

Level 21 12 3 4 5 6 78 9 10 11 12 13 14 15 16 17 18

Level 22 12 3 4 5 6 78 9 10 11 12 13 14 15 16 17 18

Appendix 11 ActivPAL instruction

Thank you for wearing the activity monitor. It records how much of the day you are sitting, standing or walking.
We would like you to wear it for at least seven days, you can take the monitor off your leg during the night, but this means attaching it to your thigh in the morning as soon as possible after you get up, and taking it off before you go to bed.
The monitor only records for 10 days starting from the time you were given the monitor so please could you wear it next day of receiving it!
After you have worn the monitor for seven days, could you please send it back to me in the self-addressed stamped envelope. We can then download the data from the monitor onto a computer and see how much of the day you spent either walking, standing or lying/sitting.

Applying the activPAL (activity monitor)
It is most comfortable to wear the monitor attached to the thigh (see picture). It should be positioned on the midline of the thigh, between the hip and the knee. However, it will function correctly if placed anywhere on the front of the thigh in the orientation indicated by the figure on the front panel. (the little man should be standing/UP pointing upwards)
The monitoring device and the tape are not waterproof and they should be removed for bathing. The skin should be thoroughly dried after bathing to maximise the adherence of the tape/gel.

Finally, please return the included form and note which days the monitor was attached to your thigh.

Please contact me on 0131 4740000 if you have any questions regarding the monitor.

Thank you very much for helping with this study,

Asfarina Zanudin
PhD student
Physiotherapy Division
School of Health Sciences
Queen Margaret University Drive
Musselburgh
EH21 6UU
Email: AZanudin@qmu.ac.uk

	Days& dates	time
Day 1		
Day 2		
Day 3		
Day 4		
Day 5		
Day 6		
Day 7		

Comments

_____
_____
_____
_____
_____
_____
_____
_____

Please fill in the day and the times above and return with the monitor in the self-addressed stamped envelope

Appendix 12 Feedback on the exercise programme questionnaire

Feedback on the Exercise Programme

1. Do you feel that the level of the exercise programme was appropriate for you?
Choose best answer

- ☐ Yes
- ☐ Usually, but sometimes too easy
- ☐ Usually, but sometimes too difficult
- ☐ No, much too easy
- ☐ No, much too difficult

2. Would you find it more enjoyable to exercise in a group or individually?
Choose best answer

- ☐ Group
- ☐ Alone
- ☐ Makes no difference

3. Do you feel that the length of the exercise session was:
Choose best answer

- ☐ Just right
- ☐ Too long, it should have only been _____ minutes long
- ☐ Too short, it should have been _____ minutes long

4. How did you feel about the variety of exercises during each individual session?
Choose best answer

- ☐ Good variety of exercises
- ☐ Not enough variety/I sometimes got bored
- ☐ Too many different exercises
- ☐ Other: _____

_____

5. How do you feel about any alterations to the sessions over the course of the 24 week programme?*Choose best answer*

- ☐ I liked how the sessions were altered as I got more used to the exercises
- ☐ The sessions were altered, but I would have rather it stayed consistent
- ☐ There were no alterations and I liked that
- ☐ There were no alterations and I would have liked some changes
- ☐ Other: _____

_____

6. Did you experience any muscle soreness/stiffness, pain, spasticity or similar adverse effects as a result of the exercise? *Please describe briefly.*

_____

_____

_____

_____

7. Do you feel that the amount of supervision/instruction that was provided was:
Choose best answer, add comments if you wish

- ☐ Just right — Comments: _____
- ☐ Too much _____
- ☐ Not enough _____

8. How did you feel about the location of the exercise programme? Where would you rather be exercising? *Choose best answer*

- ☐ Local leisure centre
- ☐ Physiotherapy clinic
- ☐ Other: _____

9. Did you notice any differences in your daily functioning since the start of the exercise programme? *Please describe briefly.*

_____

_____

_____

_____

10. Would you consider continuing the exercise programme on your own now that the study is over? *Choose best answer*

☐ Yes ☐ No

☐ Yes, but only for 1-2 sessions per week ☐ Other: _____

11. If you are considering continuing the exercise programme which components would you include? *Check all that apply*

☐ Strength circuit ☐ Stretching/flexibility

☐ Aerobic component ☐ All components

12. If you are considering continuing the exercise programme where do you think you would be exercising? *Choose best answer*

☐ I'd like to continue going to my local leisure centre ☐ Don't know

☐ I will modify the programme to do as much as possible at home ☐ Other: _____

13. Please indicate any non-gym-based physical activities (from your initial exercise questionnaire) you plan to continue participating in?

☐ Activity 1: ..... (*to be filled in by researcher)*

☐ Activity 2: ..... (*to be filled in by researcher)*

☐ Activity 3: ..... (*to be filled in by researcher)*

☐ I was not participating in any other physical activities *(Go to question 14)*

If applicable, please state the reason you are no longer participating in the activity listed above:

_____

_____

14. Have you started any new activities since the beginning of the exercise programme:

☐ Yes: _____ ☐ No *(Go to question 15)*

If yes, do you plan to continue participating in these activities?

☐ Yes ☐ No, - Why not? _____

15. Please leave any additional comments you may have about the exercise programme , participation in other physical activities, and/or participation in this study.

_____

_____

_____

_____

_____

_____

_____

Thank you for your cooperation in the study!

Appendix 13 Participant information sheet

Queen Margaret University

EDINBURGH

Information sheet

Part 1

Study Title: **The effects of a community exercise programme on young people with Cerebral Palsy**.

I am looking for volunteers to participate in this research study. This Information sheet is given to you because you have been invited to take part in the study. I feel it is quite important for you to know about the study before you decide. Please read this information sheet carefully and talk to others about the study if you wish.

- Part 1 Explains the purpose of the study and what will happen if you decide to take part
- Part 2 Gives you more detailed information about the conduct of the study

If you have any questions, you can talk to us. Take your time to decide whether or not you wish to take part.

What is the purpose of the study?
We want to study how a community-based exercise programme will affect function, physical fitness, physical activity levels, self-esteem, and quality of life in young adults and adolescents who have Cerebral Palsy.

Why have I been invited?
You have been invited because you have Cerebral Palsy, are the right age (16-25 years), have been discharged from paediatric service, and are not currently regularly (i.e. twice a week or more) exercising in a gym.

We need a group of people to either take part in the exercise programme or be part of the control group. Which group you will be part of is entirely decided by chance, ('names out of a hat'). If you are assigned to the control group you won't go through the exercise programme

initially, but we will compare your function, fitness, activity level, etc to people who have done the exercise training. This will allow us to determine the effects of exercise training.

Do I have to take part?
It is entirely up to you to decide whether or not to take part in this study. You are not required to give a reason if you decide not to take part. If you do decide to take part, you are free to withdraw from the study at any time without giving a reason. If you do withdraw from the study the data collected during any previous assessment visits will be retained. If you decide to take part, you will be asked to sign a consent form.

What will happen if I take part?
You will be assigned to either the group that is doing the exercise programme or the control group.

If you are in the exercise group, you will take part in an 18-week exercise programme. The exercise training will take place at your local gym. For the first session and between 2-4 other sessions, an experienced physiotherapist will come to the gym with you and instruct you on the circuit exercise programme which will be adapted to your ability and needs. For the other sessions a fitness instructor from the gym will work with you to complete the circuit programme, which combines leg strength, core stability, endurance/aerobic, and flexibility exercises. For the first 6 weeks you will be expected to attend 3 supervised sessions per week. During weeks 7-12 you will attend 2 supervised sessions per week and will be encouraged to complete 1 more session on your own per week. In the last 6 weeks (13-18) there is no prescribed number of sessions but you will be encouraged to complete 3 sessions per week on your own. The times and day of the training sessions are entirely up to you, as long as the physiotherapist is able to accompany you to the gym on several occasions.

If you are in the control group you will not take part in the exercise programme. You will, however, be given free leisure centre access for 5 months once the study is complete.

Regardless of which group you are in, everyone who participates in the study will undergo four assessment sessions at the Queen Margaret University motion analysis laboratory on days and times which suit you. You will be reimbursed for travel to and from the University (which will include travel by taxi within Edinburgh). Each assessment will take about 1.5 to 2 hours to complete. These will take place before the first training session (or in the case of the control group shortly after you enter the study) and at 6, 12, and 18 weeks after the start of the study.

You will be asked to wear shorts and a t-shirt for each assessment. During the assessment a number of different measures will be recorded. You will be asked to perform activities such as walking, picking items from the floor, getting up from sitting etc. Your muscle strength will be measured for a number of muscle groups in your legs. The flexibility of your ankle, knee and hip will be recorded. You will also undergo gait analysis. During this portion of the assessment, reflective balls (called markers) will be stuck to your legs and hips and you will be asked to walk

back and forth in the lab. The markers will allow us to record your movement patterns in three dimensions using multiple cameras linked to a computer.

During the first and last assessment you will be asked to complete four short questionnaires.

Before the exercise programme begins you will be given an activity monitor to wear for 7 days. The monitor is about the size of a credit card and is attached to the front of your thigh. It will record information about how many steps you take as well as the time you spend walking, standing, and sitting or lying down.

Lastly, those in the exercise group will be asked to keep a log of your gym attendance and any muscle soreness or other discomfort you feel during or after your sessions. Although, some initial muscle soreness is normal for people when starting an exercise programme, you should inform your physiotherapist if this soreness continues or if you experience any other pain or discomfort as a result of the exercise programme and he/she will then modify your exercise programme appropriately.

What are the possible disadvantages and risks of taking part?
The assessment procedures involved are safe and common clinical assessments. However, there are minimal risks such as the possibility to fall, trip etc. The exercise programme will be tailored to your abilities and needs and will be supervised by a fitness instructor who has been instructed by an experienced physiotherapist so that risk of injury as a result of the exercise programme will be minimal.

What are the possible benefits of taking part?
By taking part in this study you will get to take part in a free supervised exercise programme either during the study if you are part of the exercise group or after the study has been completed if you are in the control group. During this time you will have unlimited access to your local community leisure centre.

There are also indirect benefits for other young adults with CP. This study will help to develop an exercise programme to assist young adults in their transition from childhood to adult health care and hopefully help improve their daily function and quality of life.

What if there is a problem?
Any complaint about the study, the way you have been dealt with during the study or any possible harm you might suffer will be addressed. Detailed information is given in part 2.

Will my taking part in the study be kept confidential?
If you agree, your general practitioner will be made aware that you are participating in the study, but all information collected about you is kept strictly confidential; details are included in part 2.

Contact Details:

Ms Asfarina Zanudin
PhD student
Queen Margaret University
Queen Margaret University Drive
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East Lothian
EH21 6UU
Tel: 0131 474 0000
E-mail: AZanudin@qmu.ac.uk

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Email: mvanderlinden@qmu.ac.uk

This completes Part 1 of the information sheet.
If the information in Part 1 has interested you and you are considering the participation, please continue to read the additional information in Part 2 before making any decision.

Part 2

What if there is a problem?
Any complaint about the study, the way you have been dealt with during the study or any possible harm you might suffer due to negligence will be addressed. Queen Margaret University has a liability insurance scheme for compensation as a result of harm caused due to the negligence on the part of the researcher in connection with the above mentioned study but no compensation arrangements are there for non-negligence harm. In the unlikely event that a participant loses capacity to consent during the study, this participant will be withdrawn from the study. Data collection during the previous assessment visits will be retained.

Will my taking part in the study be kept confidential?
Only your GP will be informed of your participation in the study, however this is not required. You can omit ticking the box in the consent from. If you do not want your GP to be informed the information collected about the participants will be kept strictly confidential. The data will be stored securely in locked cabinets or on a password protected network at Queen Margaret University. Every participant is given a code at the beginning of the study and only this code, not your name, is associated with any recorded data. Care is taken to remove any information that could identify you from any information presented, published or taken out of the university for any reason.

The data will be accessed only by researchers involved in the study and the research committee responsible for monitoring the quality of research.

What will happen to the results of the research study?

The results of the study will be submitted for publication in scientific journals. Care will be taken that the participants are not identifiable in any of the materials published and all the data collected will be kept for 10 years and will then be disposed of carefully.

Who is organising and funding the research?
The study is funded by The James and Grace Anderson Trust and 'La Fondation Motrice'. Asfarina Zanudin is a PhD student at Queen Margaret University, will coordinate the study under the supervision of Dr Marietta van der Linden, a senior research fellow at Queen Margaret University.

Who has reviewed the study?
This study was given a favourable ethical opinion by the South East Scotland Research Ethical Committee and the Queen Margaret University Research Ethics Committee.

Thank you for your valuable time. If you have any questions you are welcome to contact Ms Asfarina Zanudin during office hours (contact details above).

If you want to talk to an independent advisor please contact Dr Frederike van Wijck for general information regarding taking part in a research study, andJacky Yirrell for information specific to this research.

Independent Contacts:

Frederike van Wijck
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Jacky Yirrell
Clinical Specialist Physiotherapist in
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If you have any complaints before, during, or after the study please contact Kim Gilchrist, Senior Research Officer at Queen Margaret University during office hours.

Kim Gilchrist
Senior Research Officer
Queen Margaret University
Queen Margaret University Drive
Musselburgh

Appendix 14 Craigalbert Leisure Centre (informal chatting)

Focus Group with 3 young people;
M - aged nearly 15y (female)
L age 18y (female)
K 19y (male)
The YP had been sent out the information sheets two weeks earlier and asked to read them and for their permission to take part in a round-table discussion about the study;
The effects of exercise therapy on young adults with cerebral palsy that is being conducted by QMU.
They were also given some time before the start of the meeting to read through the information sheet again. The meeting consisted of an informal style conversation between themselves and facilitated by Irene Croal, PLT at SCCMI, using the questionnaire provided by QMU.
The questionnaires are enclosed, completed by each participant and additional notes provided here.

- K commented that he was already a convert to exercise as he finds that flexibility exercises help to improve his ROM just as it does in able bodied people. He finds that when he does not do his training for a few weeks, he begins to lose his balance more when travelling on the bus, due to loss of core strength.
- Sometimes K finds that with other commitments – he attends college 4 days a week that he is less inclined to attend the gym. This study might provide the added incentive that he needs
- M said that because the information sheet mentions *pain and discomfort* that that might be enough to put some people off.
- M said that she does experience pain after intense physiotherapy sessions, but it does not put her off as she knows it has been good for her
- L said that she experiences quite a lot of back pain and she thinks that exercise can make it worse and it sometimes puts her off exercising
- K said that attending QMU 4 times would put him off because he doesn't know if it would mean taking time off college or how he would get there. Would the times be to suit him or at times that may co-incide with college. Because he would not know the answer to these questions, that he says might be enough to put him off responding.
- M said that she would be worried about getting to QMU and they should say in the information sheet how you would get there and when exactly they would have to attend
- K already does a lot of fitness training and was concerned that if he took part that he would have to stop his regular training
- K and M discussed the description in the information sheet about the types of exercise and the measurements made and thought there was an over-emphasis on legs. They wanted to know if the upper limbs would be targeted more if they wanted them to be.
- One participant , was very concerned by the fact that their participation on the trial would be shared with their GP. The concern was that as a recipient of disability living allowance, they are required to have a medical and so if the doctor thinks that they can take part in a 6 month exercise programme, then they should be able to work and have their DLA withdrawn.